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CHILDHOOD CANCER AS CHRONIC DISEASE: THE IMPACT ON SIBLINGS

by

(C) KATHLEEN MARIE BRETT

A THESIS
SUBMITTED TO THE FACULTY OF GRADUATE STUDIES AND RESEARCH
IN PARTIAL FULFILMENT OF THE REQUIREMENTS FOR THE DEGREE
OF MASTER OF NURSING

FACULTY OF NURSING

EDMONTON, ALBERTA
SPRING 1987

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Supervisor

Date Dec. 10, 1986

Abstract

This study examined the coping responses of siblings (and parents) of long-term survivors of childhood leukemia. Ten siblings, four long-term survivors, and seven parents from four families who identified themselves as coping "well" or "fairly well" with the experience were interviewed, following Glaser and Strauss' (1967) grounded theory approach. The inter-related processes of parental and sibling situational appraisal, reappraisal, and coping strategy use were investigated. "Balancing the demands" between normalization and protection concerns was identified as the core conceptual category which defined the parental coping process. The "balancing the demands" decision was determined by the parents' appraisals of the leukemia's implications for their ill children and families, including their sense of control over the disease outcome. Important variations between parental and sibling situational re-appraisals were noted, and related to four unique characteristics of sibling appraisal. Parental coping strategies were found to include both problem-focussed and emotion-focussed responses, while sibling coping was almost exclusively emotion-focussed. "Not talking about it" was the most widely used emotion-focussed coping strategy by both parents and siblings. A variety of long-term coping strategy outcomes for siblings were identified. These included overall limited anxiety about the disease and no obvious signs of psychosocial maladaptation, as well as minimal knowledge about leukemia. Various implications of the study findings for nursing research and clinical practice with families of pediatric oncology patients were discussed.

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Chapter 1

Introduction

Statement of the Problem

Dramatic advances in therapeutic techniques in the past few decades have changed the typical course of childhood cancer from an acute and usually fatal one to a long-term process of uncertain outcome (Pfefferbaum, 1980; Van Eys, 1977, 1981). Research on family coping with other long-term childhood diseases and handicaps demonstrates that siblings often develop a variety of maladaptive responses to chronic illness or disability (i.e. Allan, Townley, & Phelan, 1974; Lavigne & Ryan, 1979; San Martino & Newman, 1974; Teitz, McSherry, & Britt, 1977; Tew & Laurence, 1973). Certain cancer researchers have suggested that siblings of long-term cancer survivors may be at more risk for future psychological problems than the patients themselves (Cairns, Clark, Smith, & Lansky, 1979; Powazek, Payne, Goff, Paulson, & Stagner, 1980; Spinetta, 1981). Nonetheless, only one study of the adaptation of siblings of long-term survivors of childhood cancer is reported in the literature (Gogan, Koocher, Foster, & O'Malley, 1977), and its results are inconclusive. At present, therefore, nurses do not know how well siblings adjust to living with chronic childhood cancer, what strategies they use to cope with the situation, nor what factors influence their adjustment. Clearly, however, this knowledge is a crucial prerequisite if nurses are to assist families to live with the modern experience of childhood cancer.

The purpose of this study was to develop an understanding of the experience of siblings of long-term survivors of childhood cancer. A qualitative approach was used in order to elucidate the fundamental issues and variables of interest from the perspective of the subjects themselves. From this information, a theory that conceptualized and explained the experience of these siblings (and their families) was developed.

Background and Rationale

The Sibling Relationship

The vast majority of children (80 to 90%) have siblings (Cicirelli, 1977; McKeever, 1983), and in most cases, these sibling ties will provide the longest-lasting relationship of the children's lives (Bank & Kahn, 1982; Siemon, 1984). The sibling relationship is an intense one which exerts a major influence on personal development. Bank and Kahn (1982, p.15) call this influence the sibling bond and describe it as "the fitting together of two people's identities, from which each gets a sense of constancy and of being a distinct individual". The existence of such a bond between siblings means that the variety of functions they serve for each other all contribute in some fashion (positive or negative) to each other's psychosocial development and self-definition (Nye & Berardo, 1966).

Considering the major impact that siblings have on each other- and on the family as a whole- it is perhaps surprising that until recently, sibling relationships have remained a relatively unstudied component of the complex unit known as the family system (Bank & Kahn, 1982; Lamb, 1982; Pfouts, 1976). While a current resurgence of interest in the topic has resulted in a considerable body of literature investigating the formative significance of sibling relationships (Lamb, 1982), siblings continue to be overlooked or only superficially acknowledged in most family health or family crisis research (Bank & Kahn, 1982; Masters, Cerreto, & Mendlowitz, 1983; Pfouts, 1976). The lack of attention paid to the special world of siblings has led to significant informational deficits in the professional literature concerned with family-centered health-care. One of these information gaps concerns the impact upon siblings and their families of living with long-term survivors of childhood cancer.

Childhood Cancer as Chronic Disease

During the 1950's and early 1960's, when the first studies were done on the family response to childhood cancer, 84% of patients with acute lymphoblastic leukemia (the most common childhood malignancy) were dead within eight weeks of the onset of their symptoms (Gaddy & Wood, 1982). Today, however, 50% of all children with cancer can expect to be cured of their disease, and most of the remainder will survive for several years (Pfefferbaum, 1980; Van Eys, 1981). The modern reality of childhood cancer is that "the disease, often curable, is at all times chronic" (Van Eys, p.37). This major change in the nature of the illness has profound implications for the families forced to live with it (Kling, 1980; Koocher & O'Malley, 1981; Spinetta, 1982; Van Eys, 1977).

Given the lack of specific information in the literature about the family response to long-term childhood cancer, it is important to consider how valid are generalizations from studies of other chronic conditions for providing insight into the impact of "cured" acute leukemia. While it is clear that different disorders of childhood place differing sorts of demands on families (Ferrari, Matthews, & Barabas, 1983; Lavigne & Ryan, 1979), several authors argue that there is a central core of family challenges common to all such conditions (Kaplan, Grobstein, & Smith, 1976; Kling, 1980; McKeever, 1983; Meyerowitz & Kaplan, 1967; Pless & Pinkerton, 1975). For example, all chronic childhood diseases or handicaps present the family with a child who is not "normal", whose very presence renders the family itself "abnormal" (Anderson, 1981). Furthermore, chronic childhood illness requires meaningful change of the family in its role structure and interaction patterns in order to maintain family equilibrium in the face of the new situational demands (Meyerowitz & Kaplan, 1967). The family response to the challenge of any chronic childhood disorder will be determined by a complex interplay of illness-related variables with factors inherent to the family unit, its individual members, and its wider social context (Ferrari et al., 1983; Pless & Pinkerton, 1975). According to Montgomery (1981), the characteristic most important in determining family response is predictability- a characteristic notable for its absence in the onset of many chronic conditions of childhood.

Childhood cancer is therefore similar in several ways to other chronic childhood conditions in its impact upon the family. Its onset is unpredictable, it alters family self-perception and interaction patterns, and its effect is largely determined by existing family strengths and weaknesses. To this extent, then, it is valid to generalize findings about sibling responses to other chronic childhood conditions to the case of long-term childhood cancer. At the same time, however, childhood cancer is unlike most other chronic conditions in its unique social stigma, uncertain prognosis, painful and often disfiguring treatments- and the fears of suffering and death it engenders in all concerned (Sontag, 1978; Van Eys, 1977). These characteristics are of sufficient importance to warrant the designation of chronic childhood cancer as a relatively unique health condition, requiring individual research attention in order to supplement the more general understanding currently available from the literature.

The Research Question

The general purpose of this study was to explore and describe the experience, for a child, of being a sibling of a long-term survivor of childhood cancer. Originally, the study was designed to answer the following specific questions:

1. How does a child describe the experience of being a sibling of a long-term survivor of childhood cancer?

2. How do the child's parents and siblings (including the long-term survivor) describe the response of the child to the situation?

3. How do the various family members describe the changing family and child responses at the various stages

of the disease and treatment?

4. What factors in the child, the family, and the situation appear to have influenced the child's response to living with chronic cancer in a sibling?

As can be expected with the flexibility of the qualitative approach, the focus of this study changed somewhat as it progressed, due to the characteristics of the sample and to the major themes which emerged from the open-ended interviewing of the subjects. The sample families all included a child who was a long-term survivor of acute lymphoblastic leukemia, and all described themselves as coping "well" or "fairly well" with the ramifications of the situation. Within these self-identified "well-coping" families, it soon became clear that the responses of the well siblings were largely determined by the perceptions and coping strategies of their parents. Specific attention, therefore, was directed towards uncovering the changing situational appraisals of both the siblings and their parents, and the implications of these appraisals for the subsequent coping strategies used by both groups.

In the end, therefore, the study retained its original purpose, but the specific research questions changed. The actual questions which this study addressed were the following:

1. How do members of families who see themselves as coping well describe the experience of living with a long-term survivor of acute lymphoblastic leukemia (from the time of symptom onset to the present), for themselves and for their families?

2. In families who define themselves as having coped well with "cured" childhood leukemia, how did the siblings' and the parents' perceptions of the disease and its implications change over time, and what factors appear to have affected these appraisals?

3. What strategies did the siblings and parents use to cope with long-term leukemia in a child, and what is the relationship between the coping strategies used by the siblings and by their parents?

4. What appear to be the long-term consequences for the siblings of sibling and parental coping strategies used in families who define themselves as coping well with the experience of longterm childhood leukemia?

Chapter 2

Review of the Literature

The long-term adjustment of the well siblings of chronically ill children has not been extensively investigated in the research on childhood cancer, although it has been studied by numerous researchers interested in other chronic childhood conditions. Therefore, for the purposes of this discussion, these two sets of literature were combined and analyzed in terms of their implications for siblings of long-term survivors of childhood cancer. The studies under review were divided into three categories according to their authors' particular theoretical perspectives. Whether clearly delineated in the research reports or merely alluded to, each of these three perspectives implies a set of assumptions about human beings and their world which have major implications for the subsequent research questions, data collection and analysis methods, and interpretation of findings.

The three major perspectives used by the researchers of sibling responses to chronic childhood disorders can be described as crisis/ stress, family systems, and coping. The first group of studies focuses upon the family stress resulting from the crisis of chronic childhood illness or handicap, and describes the psychological problems of individual family members produced by such stress. The second group of studies uses a family systems approach to examine the impact of chronic illness on overall family functioning. The third group of studies stresses the notion of positive adaptation and examines the strategies used by families to cope with chronic childhood disorders.

Crisis / Stress

This section examines the studies of the family response to long-term childhood illness or handicap where the terms "crisis" and "stress" were used as conceptual paradigms about illness and the family, and not merely as descriptive terms. According to Gayton, Friedman, Tavormina, and Tucker (1977), the major assumption of the crisis/ stress paradigm is that the family stress engendered by the onset of chronic childhood illness will almost invariably result in psychopathology for the individual family members, including the siblings. This perspective is the most frequently used in the literature on the sibling response to chronic childhood disease and handicap. It is especially common in the older research, which may be partially a reflection of the grimmer prognoses and more limited treatment options that were available for many childhood conditions only a few decades ago.

The crisis/ stress perspective has been used in studies related to (among others) cystic fibrosis (Allan et al., 1974; Turk, 1964), congenital heart disease

(Apley, Barbour, & Westmacott, 1967), mental retardation (Gath, 1972; Holt, 1958; Kaplan, 1969; San Martino & Newman, 1974), spina bifida (Tew & Laurence, 1973), and childhood cancer (Binger, Albin, Feuerstein, Kusher, Zoger, & Mikkelsen, 1969; Cain, Fast, & Erickson, 1964; Cairns et al., 1979; Gogan et al., 1977; Kaplan et al., 1976; Peck, 1979; Spinetta, 1981; Teitz et al., 1977; Wold & Townes, 1969). These studies can be divided into two groups ("early" and "later"), according to their relative sophistication. Most of the early studies in this area (carried out largely from the late 1950's to early 1970's) used simplistic data collection and analysis methods. The later crisis/ stress studies of the sibling response to chronic childhood illness tended to be more methodologically sound and theoretically complex.

The psychopathology assumption implicit in the crisis/ stress paradigm could be described as the A (crisis) leads to X (psychopathology) theory of illness (Young, 1983; Longo & Bond, 1984). For Simeonsson & McHale (1981, p. 157), this postulation of probable maladjustment is a reflection of our society's cultural belief that an ill or handicapped child represents "a burden- a misfortune that the family must bear". A major implication of this belief is that most of the studies in this category focused primarily upon the incidence and types of sibling (or family) maladjustment- and/or upon those situational and personal factors associated with such maladjustment. Kazak and Marvin (1984) point out that researchers using this paradigm typically failed to investigate positive coping strategies and styles. Furthermore, most authors using the crisis/ stress perspective included few suggestions for preventing or minimizing those negative repercussions of chronic illness upon siblings which they appeared to have regarded as virtually inevitable.

The "early" crisis/ stress studies are notable for their numerous methodological weaknesses, including the lack of representative sampling, statistical tests of significance, and consideration of influencing variables and alternate theoretical explanations. Furthermore, because of their reliance upon the one-time parental interview and chart review data collection techniques, the "early" researchers did not examine the siblings' own perceptions of their situations. Not surprisingly, the results obtained by these researchers tended to bear out the fundamental assumptions of their crisis/ stress perspective. Their major finding was that chronic illness or handicap in children tends to be associated with relatively high levels of psychosomatic, emotional, behavioural, and academic problems amongst their siblings.

Of the sibling responses to chronic childhood disorders listed in Appendix A, those noted most frequently by the early crisis/ stress researchers were jealousy and resentment (Allan et al., 1974; Binger et al., 1969; Turk, 1964), and/ or increased anxiety and decreased self-esteem (Cain et al., 1964; San Martino & Newman, 1974). Some study findings suggested that these feelings tended to be "acted out" through externalizing behaviours such as decreased school performance (Binger et al., 1969; Blinder, 1972; Cain et al., 1964), increased fighting and aggressiveness (Blinder, 1972), and social withdrawal (Blinder, 1972; Cain et al., 1964; Poznanski, 1973; Wold & Townes, 1969). They could also be internalized, resulting in psychosomatic illnesses (Binger et al., 1969; Blinder, 1972; Turk, 1964; Wold & Townes, 1969), depression (Binger et al., 1969; Cain et al., 1964; San Martino & Newman, 1974), and a variety of other psychological disturbances.

"Later" users of the crisis/stress perspective on the sibling response to chronic childhood disorders often had findings similar to their predecessors, but they attempted to strengthen the validity of these results in several ways. Larger sample sizes were used, and control and comparison groups were included. A variety of research methods were employed and statistical tests of significance were utilized in the analysis of findings. Interestingly, the results of these more sophisticated studies were less unanimous than those of their predecessors. For example, Lavigne and Ryan (1979) and Tew and Laurence (1973) found siblings of children with various types of chronic conditions to demonstrate significantly more emotional maladjustment than children with healthy siblings. In contrast, both Gath (1972) and Breslau, Weitzman, and Messenger (1981) found no significant differences in the overall amount of behavioural disturbance demonstrated by siblings of handicapped versus well children. However, two cancer-oriented studies that used the patients themselves as a control group found that the well siblings fared as badly or worse than their ill brothers or sisters in overall psychological adaptation (Cairns et al., 1979; Spinetta, 1981). In the only study related to siblings of longterm cancer survivors (Gogan et al., 1977), brief case studies were presented to demonstrate negative ongoing effects, but no attempt was made to estimate the overall frequency of such responses nor the factors contributing to them.

A variety of possible influencing variables on the sibling response to chronic childhood disorders were investigated by the later users of the crisis/ stress perspective, suggesting a more complex conceptualization of the problem than the original A (crisis) produces X (psychopathology) formulation. These variables can be classified according to whether they pertained to the family, the ill child, the well sibling, or the disease (as outlined in Appendix B). Only two studies using the crisis/ stress perspective examined specific family-related variables. In a study of the impact of culture, Teitz et al. (1977) found that Mexican-Americans assigned fundamentally different meanings to the experience of chronic illness and family crisis than did Anglo-Americans. According to Farber (1960), such differences in perception were partially attributable to the impact of socioeconomic status. Farber demonstrated that while lower socioeconomic status families responded primarily to the organizational and financial "burden of care" which chronic illness represented, the experience of families of higher socioeconomic status was dominated by the shattered expectations accompanying their loss of a "normal" child.

The research examining the response-determining variables related to the ill child, the well sibling, and the disease has resulted in a range of widely varied findings that precludes definitive conclusions. For example, Lavigne and Ryan (1979) found that female siblings younger and male siblings older than the ill child responded most poorly to the chronic illness experience. The results of Breslau et al. (1981), however, showed older sisters and younger brothers to be experiencing the most maladjustment. Similarly, while Tew and Laurence (1973) found sibling adjustment to be significantly related to the degree of handicap in the ill child, Breslau et al. (1981) and Lavigne and Ryan (1979) found no relationship between the degree of handicap and the level of sibling maladjustment. Simeonsson and McHale (1981) summarized the discrepant findings on factors influencing the sibling response to chronic childhood handicaps in a major review article. While arguing that any generalizations across studies must be tentative at best, they suggested that a typical profile of a sibling at risk for maladjustment was an oldest or younger sister of a severely handicapped boy in a small family of either low or high socioeconomic status.

An important question not dealt with by Simeonsson and McHale (1981) is how the type of chronic childhood condition affected the adjustment of the well siblings. Lavigne and Ryan (1979) and Gath (1972) argued that the characteristics and imposed circumstances associated with certain chronic childhood conditions strongly influenced the response of family members to these conditions. Disease-related characteristics cited by various investigators which appeared to affect sibling response included the visibility of the condition (Lavigne & Ryan, 1979; Poznanski, 1973), the social stigma and/or myths associated with the condition (Allan et al., 1974; Farber & Jenne, 1963; Katz, 1980; Turk, 1964), the presence of a poor prognosis (Allan et al., 1974; Burton, 1975; Peck, 1979; Turk, 1964; Wold & Townes, 1969), and the amount of parental time and attention required by the ill child (Allan et al., 1974; Burton, 1975; Kramer & Moore, 1983; Lansky, Stephenson, Weller, & Cairns, 1982; Peck, 1979; Turk, 1964; Wold & Townes, 1969). The finding that sibling adjustment may be partially determined by the type of chronic condition of the ill child indicates that caution must be taken against over-generalizing study results obtained from investigations of widely varied chronic conditions. It also supports the strategy of separate study of relatively unique conditions, such as "cured" childhood leukemia.

In summary, the users of the crisis/ stress perspective agreed that chronic childhood disorders were likely to result in a variety of adjustment problems in affected siblings. This conclusion implies that these children are thereby in need of nursing attention, both in the clinical and research realms. The users of the crisis/ stress perspective provide direction for practical and investigative involvement with affected siblings, through their elicitation of factors which may affect a child's response to living with a chronically ill or handicapped brother or sister.

Family Systems

A less common but theoretically important perspective on the sibling response to chronic childhood illness is that which views the family as a system. The most fundamental assumptions of this perspective are that the family is an entity greater than the sum of its individual members, and that a change which affects one member of the family system is bound to affect all (Bubolz & Whiren, 1984). This systems focus naturally produces different research questions, methods, and interpretations than the crisis/ stress paradigm previously discussed. The family systems component of the relevant research literature concentrates upon the investigation of family interaction, rather than upon individual psychopathology (Desmond, 1980; Longo & Bond, 1984). This orientation necessitates the use of such research techniques as family observation, family assessment, and interviews with multiple family members. However, true family analysis was not achieved in most of the studies under consideration, as fathers (and to a lesser extent siblings) were often excluded from the data collection phase.

The results of the family systems oriented studies on the response to chronic childhood illness or handicap are presented at both the system (family) and subsystem (individual) levels. At the family level, reductions in overall satisfaction levels were noted after the onset of cystic fibrosis (Gayton et al., 1977). Ferrari et al. (1983), Meyerowitz and Kaplan (1967), and Ritchie (1981) found that living with cystic fibrosis or childhood epilepsy tended to produce marked (and often dysfunctional) changes in family role patterns and expectations. O'Connor and Stachowiak (1971), on the other hand, concluded that the interaction patterns of families of mentally retarded children remained functional despite their differences from the control group of "normal" families. In studies related to the impact on individual family members of childhood kidney disease, cancer, and cystic fibrosis (respectively), Klein (1976), Sourkes (1980), and Burton (1975) noted that well siblings (especially females) were expected to take on increased responsibilities in the form of household maintenance tasks, childcare, and parent support. At the same time, however, expectations of the ill child tended to be drastically reduced.

According to Crain, Sussman, and Weil (1966), the mother-child relationship enjoyed by diabetic children was much warmer and closer than that experienced by their well siblings. Meyerowitz and Kaplan (1967) explained this finding by postulating that in the effort to re-structure family interaction patterns to accommodate the new role demands associated with chronic illness, the family often became "slowed up in its affectational and emotion-satisfying performances" with the non-ill members. Various authors noted that problems in the individual adjustment of well siblings tended to follow disruptions in the family's supportive and integrative functioning (Crain et al.; Graliker, Fishler, and Koch, 1962). In fact, Meyerowitz et al. and Crain et al. both concluded that chronic childhood illness may be more psychologically damaging to the well siblings than to the ill child. This finding concurred with the results of Spinetta (1981) and Cairns et al. (1979), who had studied the impact of childhood cancer from the crisis/ stress perspective.

The family systems perspective offers two distinct contributions to the understanding of the sibling adjustment to chronic childhood illness. First, the use of a systems framework promotes a more complex analysis of coping responses than the A leads to X formulation described earlier (Longo & Bond, 1984). Second, the multi-faceted systems perspective includes an acknowledgement of the major role played by individual appraisal in determining family member response to stressful situations (Walker, 1985). In an effort to tie together the multiple facets of the family response to chronic childhood illness, several family-systems oriented researchers have used the stress response model of Hill (1958), where A (stressor event characteristics) interacts with B (family resources) and C (the family definition of the event) to produce X (the family crisis). Hill's model suggests that no single event causes a certain family or sibling response. Instead, as Montgomery (1981) explains, crisis or stress response is the result of a dynamically interactive "fit" between individual, family, situational, and societal characteristics. In summary, then, the use of systems theory has served to broaden the investigation of the family response to chronic childhood disorders by directing researchers' attention to the complex inter-relationships between factors and persons which must be considered for meaningful analysis.

Coping

Because its focus is upon the context in which stress responses take place, the family systems framework can be combined with either the crisis/ stress perspective or the coping one to be discussed in this section. While the coping perspective is widely used in the more recent studies of the family/ sibling response to childhood illness, theoretical definitions of coping can vary significantly. Futterman & Hoffman (1973), Holaday (1984), and Venters (1981) defined coping as those strategies used by families (especially parents) to manage stress and maintain emotional and interpersonal equilibrium. Spinetta (1982), however, argued that this definition focused too narrowly upon the tasks of life and ignored the further dimension of potential growth and "the joy of living". For the purposes of this discussion, the coping perspective will be defined as that orientation which emphasizes positive family (and individual) adaptation (to the point of either equilibrium or further growth) in the presence of a chronically ill child. The studies using this perspective focussed upon the various aspects of coping with chronic childhood disorders, including the nature of coping (Barsch, 1968; Desmond, 1980; Kazak & Marvin, 1984; Kupst & Schulman, 1980; Ross-Alaolmolki, 1986), its typical course over time (Futterman & Hoffman, 1973; Obetz, Swenson, McCarthy, Gilchrist, & Burgert, 1980; Pinyerd, 1983; Venters, 1981), and associated strategies (Koch-Hattem, 1986; Krulik, 1980; Powazek et al., 1980; Spinetta & Maloney, 1978; Venters, 1981).

While users of the coping paradigm acknowledged that chronic illness can represent a major stressor for a family, their findings indicated that, on the whole, families adjusted satisfactorily or even well to chronic childhood illness (Barsch, 1968; Futterman & Hoffman, 1973; Gayton et al., 1977; Kazak & Marvin, 1984; Powazek et al., 1980; Venters, 1981). From this perspective, then, individual psychopathology is not an inevitable outcome of chronic childhood illness (Gayton et al., 1977; Klein, 1976; Schulman, 1983). For example, Cleveland and Miller (1977) reported that the siblings they investigated typically adapted well to the situation, demonstrating increased levels of maturity, altruism, and tolerance. Futterman and Hoffman (1973, p. 129) concluded that chronic childhood illness could produce "emotional growth, family cohesiveness, (and a) positive redefinition of values" within the affected families. Several authors (Desmond, 1980; Futterman & Hoffman, 1973; Graliker et al., 1962; Kupst & Schulman, 1980) explained this apparent contradiction with the crisis/ stress results by pointing out that family or individual responses which appear pathological when viewed in isolation can often be seen as adaptive when considered in context. These investigators therefore stressed that it is inappropriate to apply the usual criteria of psychopathology to families of seriously or chronically ill children.

The investigators of family coping with chronic childhood illness relied largely upon the techniques of family observation and detailed repeat interviews with family members (usually parents) in order to uncover the nature and typical course of this adaptation. Schulman (1983) and Schwirian (1976) found no pattern of relationships between variables such as family socioeconomic status or size, or the ill child's age or sex, and effective coping outcomes. However, Obetz et al. (1980), Spinetta and Maloney (1978), and Venters (1981) noted a relationship between the use of certain types of coping strategies and optimal long-term family adjustment. Taken together, the lists of successful coping strategies compiled by the researchers of the family response to chronic childhood illness are similar and include five main strategies. These strategies are the use of social support (Holaday, 1984; Kazak & Marvin, 1984; Krulik, 1980; Schilling, Gilchrist, & Schinke, 1984; Schulman, 1983; Spinetta & Maloney, 1978; Venters, 1981), achieving mastery (Futterman & Hoffman, 1973; Holaday, 1984; Iles, 1979; Krulik, 1980), normalizing the situation for the family (Anderson, 1981; Anderson & Chung, 1982; Futterman & Hoffman, 1973; Graliker et al., 1962; Holaday, 1984; Krulik, 1980; Schulman, 1983), assigning meaning to the experience (Futterman & Hoffman, 1973; Spinetta, 1981; Venters, 1981), and the use of open family communication (DelCampo, Chase, & DelCampo, 1984; Krulik, 1980; Schulman, 1983; Spinetta & Maloney, 1978).

Interestingly, despite their supposed focus upon _family_ adaptation, several of the coping-oriented studies omitted siblings from their investigations altogether (Futterman & Hoffman, 1973; Kazak & Marvin, 1984; Kupst & Schulman, 1980; Schilling et al., 1984), or dealt with them only indirectly (DelCampo et al., 1984; Obetz et al., 1980; Stebhens & Lascari, 1974). To-date, only one investigator (Koch-Hattem, 1986) has specifically examined the strategies used by siblings to cope with chronic health disorders (in this case, cancer) in a brother or sister. Major gaps therefore exist in the research literature about how siblings typically cope with chronic childhood illness, and how these strategies are related to parental coping approaches. The present study is designed to address these issues in the specific context of "cured" childhood leukemia.

Summary

Several major reviewers (Longo & Bond, 1984; Masters, Cerreto, and Mendlowitz, 1983; McKeever, 1983; Simeonsson & McHale, 1981) have concluded that wide-spread methodological deficiencies in the relevant research literature make it impossible to draw definitive conclusions about the sibling (and family) response to chronic childhood illness or handicap. Many of the studies in this area were based upon anecdotal and impressionistic information, utilizing small sample sizes and tools and techniques of undemonstrated reliability and validity. Typically, siblings were either ignored or described solely through parent report- rarely were they allowed to speak for themselves. Research findings tended to be consistent with the particular world-view of the author, and were not systematically replicated or followed up by other researchers. Despite these weaknesses, however, several tentative conclusions can be drawn from the literature which have implications for further research in the area. First, it is clear that children with chronically ill or handicapped brothers or sisters are a population potentially at risk for problems of long-term maladjustment. Second, one would expect a variety of sibling responses to chronic illness (such as those outlined in Appendix A), which would be affected by a range of inter-acting variables (such as those outlined in Appendix B).

Because so little verified information about the adjustment of siblings of long-term survivors of childhood leukemia exists, the study was designed as a beginning, factor-searching analysis of the phenomenon (Diers, 1979). In order to avoid biasing the study results through major a priori assumptions about the situation (such as those described in the foregoing review), a qualitative approach which grounded the study in the reality of the sample families was chosen.

Chapter 3

Methods

Qualitative research involves the use of induction to extract meaning from data and generate theories (Swanson & Chenitz, 1982). The particular qualitative method deemed to best meet the purposes of this study was Glaser and Strauss' (1967) "grounded theory" technique. As explained by Field and Morse (1985, p.111), the purpose of grounded theory is to "gain a fuller understanding of what constitutes reality for the participants in a particular real-life setting".

The Sample

A nonprobability sampling approach was used in this study because of its appropriateness for the qualitative research purpose of deep understanding of phenomena (Field & Morse, 1985; Bogdan & Biklen, 1982). Morse (1986), Glaser and Strauss (1970), and McCall and Simmons (1969) all suggest that the most useful non-probability technique for obtaining complete and relevant information is the theoretical (or purposive) sampling method. Using this approach, subjects are selected according to what the researcher needs to know (Morse), and more time is spent with those who turn out to be "good" informants (i.e. are co-operative, insightful, and knowledgeable about the phenomenon) (Turner, 1981). A theoretical sample of local families was therefore selected for use in this project.

For the purposes of this study, "family" was defined as the nuclear family unit, including mother, father, sisters, and brothers. While true purposive sampling requires the researcher to be familiar with all potential subjects before selecting some to interview at length (Morse, 1986), accessibility issues determined instead that initial selection be made in conjunction with the Nurse Counselor of the local pediatric oncology clinic. The initial selection criteria were the following:

1. All family members could be appropriately described as Anglo-Canadian;
2. The family includes one child who:
    a. was diagnosed with any form of childhood leukemia three or more years ago (but not more than ten years ago), OR, (if there are inadequate numbers of long-term survivors of leukemia), was diagnosed with any form of cancer three or more years ago (but not more than ten years ago), and
    b. is currently in remission, and
    c. is off active treatment for cancer and is considered cured;
3. The family includes a second child who is age ten or over and not suffering from any major handicapping or life-threatening health condition; and

4. The family lives within thirty miles of Edmonton.

Upon examining the files of the pediatric oncology clinic with the Nurse Counselor, it became obvious that the number of long-term cancer survivors was too small to allow fulfillment of all the above-listed selection criteria. When it was realized that only a minority of potential subject families could be described as Anglo-Canadian, (due to the large local immigrant and ethnic populations), selection criteria #1 was eliminated. In order to reduce other sources of potential variation between families, it was decided to focus strictly upon those with a child who had survived acute lymphoblastic leukemia. (One rationale for this decision was the finding by researchers such as Sourkes (1980) and Teitz et al. (1977) that there are significant differences in experience and coping strategies between families where a child has a solid cancer requiring surgical removal and those whose child has leukemia.) In order to boost the number of potential families with well siblings ages ten or over, selection criteria #4 was eliminated, and the maximum span from time of diagnosis was increased to twelve years. (In one case, the minimum age limit was lowered to nine and one-half years.) With these adjustments, eight families were eligible for participation in the study.

The Nurse Counselor of the pediatric oncology clinic made initial contact with each of the eight potential subject families. She briefly explained the study to them, emphasizing that participation was strictly voluntary. She then requested permission for their phone numbers to be given to the investigator. Two families refused to grant this permission, due to the unwillingness of at least one member in each family to discuss the leukemia. The investigator contacted the remaining six families. The study was described, the consent procedure was outlined, and questions were answered. Two further families were eliminated as potential subjects at this time. In one case, a father would not permit the words cancer or leukemia to be used with his children. In the other case, the investigator felt that the family's current state of crisis made their participation inappropriate.

After the initial phone contact, letters were sent to the remaining four potential subject families. These letters included sample consent forms and an information sheet describing the proposed study in some detail. (Examples of these materials are contained in Appendix C.) After approximately ten days, the investigator once again telephoned each family. All parents gave verbal consent to participate in the study, and interviews were then scheduled at mutually convenient times and places.

In-home interviews were conducted with three of the four families, while the fourth family preferred being interviewed at the site of the pediatric oncology clinic.

All members of the subject families ages nine and one-half and over were interviewed (except for one father who was unavailable due to work constraints). The total number of individual subjects was four mothers, three fathers, four long-term survivors, and ten siblings. Because of the small size of the study sample and the strictly voluntary method of participation, it could be criticized as being unrepresentative and potentially biased. In practical terms, however, the population from which this sample is drawn is itself very limited in size. More importantly, though, it must be remembered that sample size and representativeness are concerns relevant primarily for quantitatitve questions about frequency, distribution, and correlations (Morse, 1986; Field & Morse, 1985). A small sample size was purposely chosen in order to permit the detailed, in-depth analysis from which theory could be developed that larger investigations could later test and expand.

Data Collection

All subjects were interviewed on three separate occasions spaced approximately one month apart. Due to time constraints and parental requests, parents were usually interviewed together. All children were interviewed individually, except in two cases where joint introductory interviews were requested by the families. Interviews ranged from twenty to seventy-five minutes in length with the children and from thirty minutes to two hours with their parents.

The in-depth interview which forms the core of the grounded theory method was particularly appropriate for this project due to the sensitive nature of the topic under study. The interviewer took steps with each subject to promote the creation of a relaxed atmosphere in order to facilitate the sharing of feelings. The semi-structured interview format meant that a core of standard questions was asked of each subject. However, these questions were all asked in an open-ended style that permitted flexibility and maximum opportunity for clarification and amplification.

Several steps were taken by the researcher during the interviews to increase reliability and validity. Key questions were repeated in several different ways and on separate occasions. Subjects were encouraged to expand upon short answers and to clarify possible ambiguities. At certain points throughout each interview, the researcher would seek to verify her understanding of each subject's answers in order to ensure "shared meaning" (Spradley, 1979; Turner, 1981). Each subject was asked questions about major points and events mentioned by other interviewees in order to compare perspectives within and amongst families. The new key questions which emerged from these important points were then entered on a tally record sheet. These sheets were used to record the dominant issues discussed in each interview and thereby ensure that the same central questions were covered with each subject. During the initial reading of the transcripts of the taped interviews, unclear, contradictory, or minimal answers were noted and elaboration was sought in subsequent conversations.

As suggested by Spradley (1979), the first interview with each subject was the most unstructured in order to permit the interviewee to freely identify those themes he/she saw as most important in his/her experience with long-term cancer. The question that opened the first interviews was "How do you think you and your family have been affected by the fact of X. having had leukemia?" After general impressions of the impact of the leukemia were obtained, each subject was asked to describe his or her memories of disease-related experiences from the initial onset to the present time. In the more structured second interview, these events were discussed again, but from the perspective of the feelings that accompanied them and the meaning they held for the participants. Individual responses to leukemia-related events were focussed upon through the asking of a series of pre-determined questions derived from the literature about family reactions to living with chronic childhood disorders. (These questions, outlined in Appendix D, were not particularly useful due to the length of uneventful time for the subject families since the onset of first remission in the survivors. The dramatic sibling responses explored by the questions in Appendix D had long ago disappeared in the study sample and memories of them were fuzzy or nonexistent.)

The third interview was a summarizing interview, in which final clarification was sought and previously overlooked questions were addressed. Each subject was asked to construct a "time-line" for the interviewer, which outlined their changing perceptions over time of the survivor's health status and the implications of this status for the family and its members. Finally, all subjects were asked to describe their feelings about the three interviews. The interviewer then explained how the interview material would be used in the final report and ensured each family had her telephone number for future reference.

In total, fifty-seven interviews were conducted. These included two family interviews, nine couple interviews, and forty-six individual interviews. Further data were collected through the use of detailed field?notes about the family visits. These notes included objective descriptions of the settings, circumstances, and participants, and reflective impressions of the interview atmosphere and the interviewees' responses and behaviour. Supplementary data were obtained through discussions with the Nurse Counselor. Her memories and perceptions of the responses of the subject families since the initial diagnosis of leukemia were used for validation purposes.

Data Analysis

Analysis of data began as soon as the first transcripts were available, and served to provide direction for later interviews. The virtually simultaneous data collection and analysis meant that there was a dialectic interaction between the two (Hutchinson, 1986), permitting the researcher to move back and forth between induction and deduction as the study progressed.

According to Glaser (1978, p. 39), in a qualitative study, "theory is gradually built up inductively from the progressive stages of analysis of the data". Following the techniques suggested by Glaser and Strauss (1967), the analysis in this study involved two major phases. These were the identification of the conceptual categories and their properties, and the discovery of the relationships among these categories. Each phase involved two stages, as described by Corbin (1986). The phase one identification of the study categories involved the stages of: (1) uncovering the initial substantive codes, followed by (2) the integrating and elaborating of these codes into theoretically more abstract categories. Phase two (the discovery of category relationships) involved the following two stages: (3) linking the categories and (4) discovering the core category.

In stage one, the analysis of the tape transcripts began with the coding of the data in as many ways as possible (as suggested by Glaser, 1978) in order to ensure that all potential underlying themes were uncovered. These early substantive codes described the emotional and behavioural reactions of the subjects and the factors which appeared to influence these responses. Examples of these codes include "not talking about it", "being rescued by the doctor", "fear and what makes it scary", and "the support of friends and family". Sample data bits from the code "equality in the family" are included in Appendix E.

The "constant comparative technique" of continually comparing data bits within and among codes was used throughout the entire data analysis. The purpose behind this technique was to "generate and plausibly suggest (without...provisionally testing) many categories, properties, and hypotheses" (Glaser and Strauss, 1967, p. 104). Ideas which arose from this process were written down in a series of ongoing memos. As more data were analyzed and the relationships between the codes became clearer, the researcher was able to move to the second level of coding.

Stage two coding of the data involved two steps. First, the initial list of codes was abridged by clustering similar ones. Next, an extensive series of memos on the similarities and differences between the abridged codes were created. Based on the thinking represented by this series of memos, the researcher was then able to further consolidate the abridged codes to form categories (shown in Table 1). As pointed out by Corbin (1986) and demonstrated in Table 2, categories move the data to a more abstract level of conceptualization. For example, the early substantive codes "keeping things the same", "being normal", and "being special or different" all fit well into the conceptually more refined category of "normalizing".

Once the categories had been obtained, further data collection and analysis was directed towards identifying their properties. Identification of these properties assisted in the transition to the third stage of data

Table 1

The Categories and Sub-Categories

1. Giving meaning-
    a. Appraisal/ re-appraisal of situation;
    b. Strategies:
        i. Having faith,
        ii. Re-defining the situation.
2. Normalizing-
    a. Keeping things the same;
    b. Maintaining equality amongst the siblings.
3. Protecting-
    a. Vigilance;
    b. Protective restrictions;
    c. Life-style changes.
4. Pulling together-
    a. For practical assistance;
    b. For emotional support.
5. Not talking about it-
    a. Keeping feelings to oneself;
    b. Not thinking about it;
    c. Limited sharing/ seeking of information.

Table 2

The Clustering of Codes into Categories

Category	Abridged Codes	Initial Codes
1.Giving meaning: a)Appraisal/ Re-appraisal	Knowing something is wrong	-The danger signs; -The early responses; -Early memories.
	Fear	-Fear and the most scary things; -Uncertainty; -Hurting; -Being worried; -Awareness of the possibility of death.
	Defining the disease	-Definition of leukemia/ cancer; -The lucky breaks; -Bad parts of the disease/ therapy; -Being sick/ not sick; -Being cured; -What made it tough.
b)Strategies: (i)Having	Trusting the health-care	-Trusting the dr.; -"Such good people";

faith	team	-Being rescued by the doctor; -Being looked after by the team.
	Searching for answers	-Advice of others; -Finding an answer; -Becoming experts.
	Hoping	-Praying; -Accepting fate; -One day at a time; -Hoping.
	Taking control	-Having control; -Not trusting the doctor; -Fighting the system; -"Playing the game".
(ii)Re-defining the situation	Seeing the positive side	-The lucky breaks; -"Not as bad as it might have been"; -Enjoying life; -Doing the "right" thing; -Good memories.
	Defining the situation	-Coming to terms; -What might have been; -Cancer "not

		important"; -Defining the experience.
2. Normalizing: a)Keeping things the same	Normalizing	-Keeping things the same; -Being no different from anyone else; -"Living as normal a life as possible"; -Teasing.
b)Maintaining equality amongst the siblings	Being different or special	-Being "special"/ different; -"Equality" in the family; -Deliberate consideration of the siblings; -Being no different from anyone else; -"Living as normal a life as possible"; -Being embarrassed.
3. Protecting:	Protecting	-Precautions;

a)Vigilance b)Protective restrictions c)Life-style changes	Changes	-Protection; -Uncertainty. -Impact on life-style; -Changes in the family; -Searching for balance.
4. Pulling together: a)Practical support	Sharing the burden	-Being tough; -Mom as primary caregiver; -Pitching in; -Organizing the family; -Being looked after by the team; -Seeking assistance.
b)Emotional support	Pulling together	-Family closeness; -Pulling together; -Parents supporting each other; -Experience with cancer/death; -Support of friends and family.

5. Not talking about it: a)Keeping feelings to oneself	Not thinking about it	-Being tough; -Keeping feelings to oneself; -Not talking about things.
b)Not thinking about it	Not thinking about it	-Being tough; -Not dwelling on it/ denial; -Not letting others get you down; -Distraction and keeping busy; -Not remembering; -Thinking about the future; -Thinking about cancer.
c)Limited sharing/ seeking of information	Communicating/ not communicating	-Talking within the family/ being open; -Not talking about things; -Evangelizing.
	Being informed/ knowing	-Being informed/ the need to know; -To tell vs. not to tell;

-Seeking information;
-Knowing what to do;
-Awareness of the possibility of death;
-Remembering;
-Ignorance/ not knowing.

analysis, which focussed upon the uncovering of the linkages between categories. In order to establish these linkages, specific questions about each category were asked in the interviews, and the constant comparison of answers within and among families, and among categories, continued. Incoming data were examined for information about causes, contexts, consequences, and conditions applicable to each of the categories under study (Glaser, 1978). Relevant research and theoretical literature was then examined in an attempt to clarify, amplify, and validate findings about the category properties and linkages.

As category linkages were being explored, diagramming of possible relationships between them (as suggested by Corbin, 1986, and Hutchinson, 1986) was used to visualize the logical flow of ideas. This exercise marked the transition to stage four, involving the identification of the study's core category. The core category "balancing the demands" incorporated the two major categories "normalizing" and "protecting", and indicated their relationships to the other three categories (especially the "appraisal" component of "giving meaning"). "Balancing the demands" was the key concept which explained how families responded to the ongoing stress of leukemia in a child.

The final step of the stage four data analysis involved refining the conceptual diagrams and validating the developing theory. At this stage, it became clear that the stress/ appraisal/ coping framework of Lazarus and Folkman (1984) fit with the study findings, and could be used to advantage in organizing and diagramming them. With this assistance, the final visual depictions of the theory (i.e. Figure 1) were formulated. Validation of the theory was sought through a final re-examination of the data and the literature, questions asked in the third interviews, and consultation with knowledgeable outsiders. These secondary informants included the Nurse Counselor at the pediatric oncology clinic, members of the thesis supervisory committee, and nurses knowledgeable about the family response to childhood leukemia.

Chapter Four

Characteristics of the Sample

A variety of demographic characteristics which could have influenced family and individual coping with long-term childhood leukemia were identified and are outlined in this chapter. To some extent, these variables help explain why the sample families appraised and responded to the cancer experience as they did. Relevant demographic characteristics of the parents, well siblings, and long-term survivors are summarized in Tables 3, 4, and 5, and described below. Throughout this paper, the families will be referred to as the A, B, C, and D families. All first names have been changed.

General Family and Parental Characteristics

The following family and parental characteristics will be discussed in this section:

1. The self-perception of coping "well";

2. Demographic characteristics;

3. Major family support sources;

4. Previous experience with cancer and/or death;

5. Organizational changes to deal with hospitalization and treatment.

Table 3
Demographic Characteristics of the Parents/Families

	Residence	Ethnic Origins	Extended Family	Religion	Number of Children	Deaths prior to Leukemia	Education	Occupation
Family A	City	Mother-French Canadian Father-from Hungary	Mother from large family*	R. Catholic Religion "quite" important	5	---	High School	Dietary Aide Plumber***
Family B	Farm	Anglo-Canadian	Mother from large family grand-parents live on farm*	Agnostic/ Atheist	3	1 (Mother's brother)	Mother-University Father-post-secondary	Teacher Farmer/ millwright

Table 3 (Cont'd)

Demographic Characteristics of the Parents/Families

	Residence	Ethnic Origins	Extended Family	Religion	Number of Children	Deaths prior to Leukemia	Education	Occupation
Family C	Small Town	Mother-Anglo-Can Father-French Canadian	Father from large family**	R.Catholic Religion "very" important	4	---	High School some post-secondary	Playschool Aide Store Manager
Family D	Farm	Mother-from England Father-French Canadian	Grand-father lives on farm*	R.Catholic Religion "quite" important	4	3 from cancer-grand-mother & 2 neighbors	High School	Farm-wife Farmer/Pipefitter

*Most or all of extended family live close by
**Some extended family live close by
***Unemployed at time of study

Self-perceptions of Coping

As noted in Chapter 1, the self-selection process undergone by the sample families modified the original focus of this study. Three of the four families who participated in the study described themselves as "coping well" with long-term leukemia, and the fourth family described themselves as coping "fairly well". This self-perception contrasts with the impression gained from the four families who did not agree to participate in the study. One mother in this second group described her family as one "where everything has gone wrong". One father refused to have the words cancer or leukemia used in his home because "it is too upsetting". In another family, the only sibling refused to participate because of ongoing resentment towards the survivor.

Of its own accord, then, this study became one that examined the perceptions and reactions of families who generally defined themselves as coping well with the experience of living with a child with long-term cancer. This fact must be kept in mind while reading the subsequent descriptions, theories, and propositions, as it is clear that only a subsection of all possible family responses to the situation was uncovered in the sample under study. It should also be noted that among the three families who defined themselves as coping "well" with the leukemia, nine or more years had passed since the diagnosis. In the family that described themselves as coping "fairly well" with the disease experience, the leukemia had been diagnosed only five years previously.

Demographic Characteristics

As outlined in Table 3, the four families who participated in this study were in many ways typical of the Western Canadian province in which they lived. Two of the four families lived on farms, one resided in a small town, and one family made their home in a major city. The eight parents in the subject families were of varied ethnic backgrounds, with three having French and one having Hungarian as their first languages. In each family, both parents were working, and the total family income was between $20,000 and $50,000. Five of the parents worked in farming or blue collar jobs, and three were employed at white collar positions. However, all the families were feeling the effects of the current recession. In both the farming families, the fathers worked in oil-related industries far from home all winter in order to supplement the family income. In one urban family, the father had recently been laid off work for a prolonged period of time.

Major Family Support Sources

The families all had a variety of support sources available to them, including each other, the health-care team, relatives, and friends. Two of these sources had especially important consequences for parental situational appraisal and coping, and are re-examined in Chapter 5. These two factors were the family's close ties and traditional values, and their varying beliefs in the power of the doctor to control the leukemia outcome.

Most of the siblings and parents described their families as "close". The marriages all appeared to be strong, and the parents each identified their spouses as having been important sources of emotional support during difficult leukemia-related times. The families tended to spend a lot of time in each other's company, and do many activities together. For example, as one father stated:

> We found that, like taking the kids out, whenever we go anywhere, we take the kids with us. We go fishing, they all come. We go down visiting someplace, they always go.

The parents expressed traditional family values and identified their children as their first priority:

> I think it's important if you're going to have children, you have to put your time in for them... I mean if a kid is interested in playing ball, they want you to be there to see what they're doing...It's

important to do those things for your kid.

The parents all had good rapport with the health-care team members at the local pediatric oncology clinic, and had consulted them on a variety of occasions for advice, information, and reassurance. Two of the sets of parents described this team in especially glowing terms. As described in Chapter 5, these parents defined the doctor, rather than themselves, as being in control of the leukemia outcome. The two other sets of parents had more doubts about the abilities of modern medicine, and expressed a more internal sense of control over disease outcome. This sense of locus of control appeared to be the determining factor behind the parents' varying "balancing the demands" decisions.

A notable characteristic of the four families was that they all had large extended family support systems providing practical and psychological assistance. These relatives had provided crucial emotional support and childcare services during the difficult early days after the leukemia diagnosis. Three of the eight parents originated from large families (between six and twelve children), and all expressed a strong belief in the importance of maintaining strong family ties. The two farming families had grand-parents living on their farms, residing in separate small homes. As well as having various relatives living close by, the families (all long-time residents of the province) had made use of a network of friends and neighbours for various types of social support. For three of the families, the Catholic church provided an additional "fairly important" to "very important" source of strength and meaning for them.

Previous Experience with Cancer or Death

Before the leukemia had been diagnosed, two of the four sample families had earlier lost an close extended family member to cancer. In one case, the family had lived with and cared for the dying grandparent. After witnessing the slow decline of this relative and the deaths of two neighbourhood children from cancer, the parents in this family responded to the diagnosis of leukemia by questioning the value of traditional medicine and seeking alternate sources of therapy to supplement their child's treatment regime.

Organizational Changes to deal with Hospitalization and Treatment

During the initial hospitalizations of their children, two of the fathers took leaves-of-absence from work either to be with their wives at the hospital or to rotate child-care duties at home with them. After the child's discharge, these two families continued to emphasize the joint participation of both parents (as much as possible) in the subsequent clinic visits. In the remaining two families, the fathers continued working throughout their child's hospitalization, and the major expectation for maintaining the hospital and clinic visits was upon the mother. During parental absences, the well siblings were looked after by relatives or neighbours. In two of the four families, arrangements for clinical and hospital visits were complicated by the fact that the mothers could not drive the car.

Well Sibling Characteristics

In this section, relevant characteristics of the well siblings (including their involvement with hospital and clinic visits) will be examined.

General Characteristics

As diagrammed in Table 4, the number of well siblings in the four families was (respectively) two, three, three, and four. None of these children suffered from any known physical or mental handicaps or illnesses. At the time of initial diagnosis, the well siblings had ranged in age from unborn to thirteen years. Their present age-range was from five to twenty-four years, meaning that two were below the minimum age established for participation in this study. The average age of the interviewed siblings (two females and eight males) was fifteen. Three were younger than the long-term leukemia survivor in their family and seven were older. All of the siblings except

Table 4

General Characteristics of the Well Siblings

	No. of Well Siblings	Age at Diagnosis (Years)	Present Age (Years)	No. Older Than Survivor	No. Younger Than Survivor
Family A	4	3 to 13	14 to 24	2 males	2 females
Family B	2	2 & 14	14 & 16	2 males	---
Family C	3*	2 to 9	7 to 14	2 males	1 female*
Family D	3*	0 to 7	5 to 16	1 male	2 males*

*1 sibling too young (5-7 years) to be interviewed for this study one lived with their parents. Three (from the same family) had graduated from high-school and were working or actively pursuing employment.

Involvement with Hospitalization/ Clinic Visits

Most of the well siblings had visited their ill brother or sister only once or not at all during their hospitalization periods. In one family, none of the siblings attended subsequent clinic visits with the ill child. In the other three families, those siblings not in school routinely accompanied the parent(s) and leukemic child on their appointments at the oncology clinic. Typically, the siblings amused themselves in the clinic play-room while their brother or sister was examined by the doctor.

Long-term Survivor Characteristics

Various characteristics of the long-term survivors will be examined in this section, including disease and therapy-related factors, their emotional responses to hospitalization and treatment, and their current health status.

General Characteristics

As outlined in Table 5, the leukemia survivors (two males and two females) were ages nine and one-half, eleven, fourteen, and nineteen. One was a youngest child,

Table 5

General Characteristics of the Long-term Survivors

	Sex	Age at Diagnosis (Years)	Present Age (Years)	Hospital-izations	Maintenance Regime	Clinic Visits/Year (Current)	Treatment Related Problems
Family A	Female	7	19	Initital only - 2 1/2 weeks	Oral Meds. + I.V. MTX* every 2 wks X 3 years	1	---
Family B	Female	1	11	4 - several days to several weeks	Oral Meds. only	1	Marked Fear of Needles
Family C	Male	4	9	Initital only - 1 month	Oral Meds. only	2	Marked Fear of L.P.'s**
Family D	Male	5	14	Initial only - 1 week	Oral Meds. + extra L.P.'s** (+ Laetrile + Herbal Medicines)***		

*I.V. MTX = Intravenous Methotrexate

**L.P.'s = Lumbar Punctures

***Bracketed treatments were not prescribed by the Clinic doctor while the remaining three were middle children. All were still in first remission, with elapsed times from initial diagnosis with acute lymphoblastic leukemia of five, nine, ten, and twelve years. The nineteen year old survivor had graduated from high school, was employed fulltime, and was still living at home.

Disease and Therapy-Related Factors

Of the four survivors studied, one was one year of age at diagnosis and three were between the ages of four and six. All had initially presented with vague symptoms of lethargy, malaise, and opportunistic infections that the parents had at first attributed to a cold or flu. When their conditions had worsened, each of the children had been taken to the local general hospital, and then transferred to the large tertiary care facility for this region of the province. The four children were then placed (and have since remained) under the care of the same oncologist, and all have been dealing with the same Nurse Counselor since their initial admission. All currently attend the same pediatric oncology clinic.

The four survivors were all initially classified as low-risk leukemics, and, according to their parents, were given prognoses of a 40 to 50% chance of "cure" or long-term survival. They were treated with similar chemotherapy protocols, although ongoing revisions of the protocols produced some differences in their therapeutic experiences. For example, one child went on to show blast cells in his cerebrospinal fluid (a negative prognostic indicator) after the induction phase of his chemotherapy. This child received more radiation and more frequent follow-up lumbar punctures than did the other three children.

During the induction phases of their chemotherapy, each child was hospitalized for a two to four week period. A prophylactic course of cranial radiation and intra-thecal methotrexate was also given. The longest term survivor received intravenous methotrexate every two weeks for three years after the induction of her remission. The other three children took oral medication only during the maintenance phase of their chemotherapy. One set of parents supplemented their child's chemotherapy with a combined Laetrile and herbal medicine regime. At three years post-remission, prescribed medication was stopped for all four survivors, and surveillance lumbar punctures of four times yearly (for one year) and twice annually (for the subsequent three years) were begun. After this period (still ongoing for one of the four subjects), clinic visits were reduced to annual nonintrusive check-ups.

Emotional Responses to Hospitalization and Treatment

Two of the four sets of parents described the initial hospitalization as an especially harrowing time for their children. These two children developed a marked fear of needles, which complicated later clinic visits. On the other hand, one of the other two survivors had such an enjoyable time during her hospital stay that she regretted leaving and decided to become a nurse when she grew up! Only one child required subsequent hospitalization. She was admitted for treatment of various infections on three occasions after her initial stay.

Current Status

From the interviews, each of the four long-term survivors appeared to be physically and emotionally well. However, several ongoing problems which may be related to the leukemia therapy continue to disturb them. One boy is self-conscious about his excess weight, while precocious physical development causes concern to one of the girls. Three of the four survivors have had to repeat a year at school, and two of the four experience serious difficulties with mathematics. These academic difficulties may be due to the central nervous system prophylaxis (cranial radiation and intrathecal chemotherapy) these children received before the age of six (Eiser, 1978; Moore, Kramer, & Albin, 1986; Moss, Nannis, & Poplack, 1981).

Summary

The characteristics described above cover many of the variables mentioned in the literature (and outlined in Appendix B) as potentially influencing sibling (and family) responses to living with long-term childhood illness. However, the small sample size and lack of quantitative measuring techniques in this study preclude the drawing of firm conclusions about the relative impact of or inter-relationships amongst the possible variables. Nonetheless, based upon the qualitative analysis of the interview data, several variables (i.e. parental locus of control, sibling exposure to evidence of disease seriousness) did appear to play a pivotal role in family (and individual) coping. These factors and their implications for situational appraisal and coping strategy use are discussed at length in the following two chapters.

Chapter Five

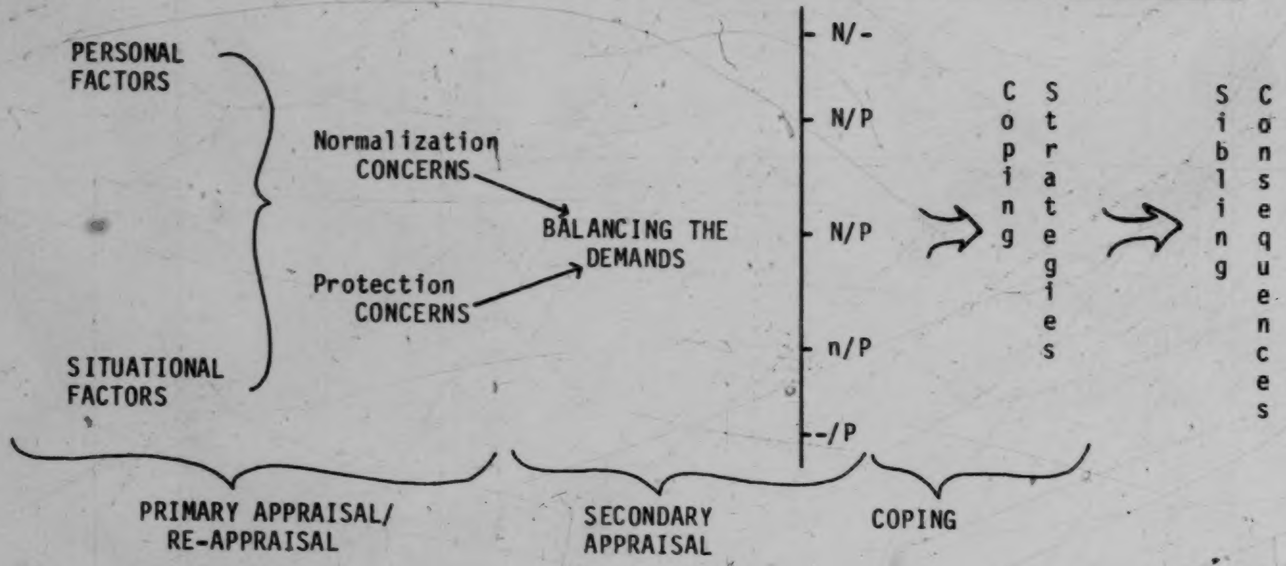
Appraisal and Re-Appraisal: Findings and Discussion

In Chapter 3, "balancing the demands" was identified as the core conceptual category which described the parents' coping responses to living with leukemia in a child. This response provided a consistent underlying theme in the families' experiences with leukemia since the time of the diagnosis, and largely determined the siblings' understanding of the situation. As depicted in Figure 1, the parents' "balancing the demands" decision was a key component of the family coping process which involved appraisals (and re-appraisals) of the implications of the leukemia, followed by varied responses to the situation based upon these perceived meanings. Appraisals of the leukemia experience (both parental and sibling) are examined in detail in this chapter. Chapter 6 focuses upon the parents' "balancing the demands" decisions, subsequent coping strategy use by both parents and siblings, and apparent long-term consequences of these strategies for the well siblings. Due to the integration of literature review into ~~the data~~ analysis methods used with grounded theory, findings and discussion will be combined in the subsequent chapters.

A definition of coping which fits the purpose of this study is: "efforts, both action oriented and intrapsychic,

Figure 1

Schematic Representation of Family Coping with Long-term Childhood Leukemia


PERSONAL FACTORS
SITUATIONAL FACTORS
Normalization CONCERNS
Protection CONCERNS
BALANCING THE DEMANDS
N/-
N/P
N/P
n/P
-/P
Coping Strategies
Sibling Consequences
PRIMARY APPRAISAL/ RE-APPRAISAL
SECONDARY APPRAISAL
COPING

Balancing the Demands:

N/- = Total emphasis on Normalization
n/P = More emphasis on Protection than Normalization
N/P = Equal emphasis on Normalization and Protection
N/p = More emphasis on Normalization than Protection
-/P = Total emphasis on Protection

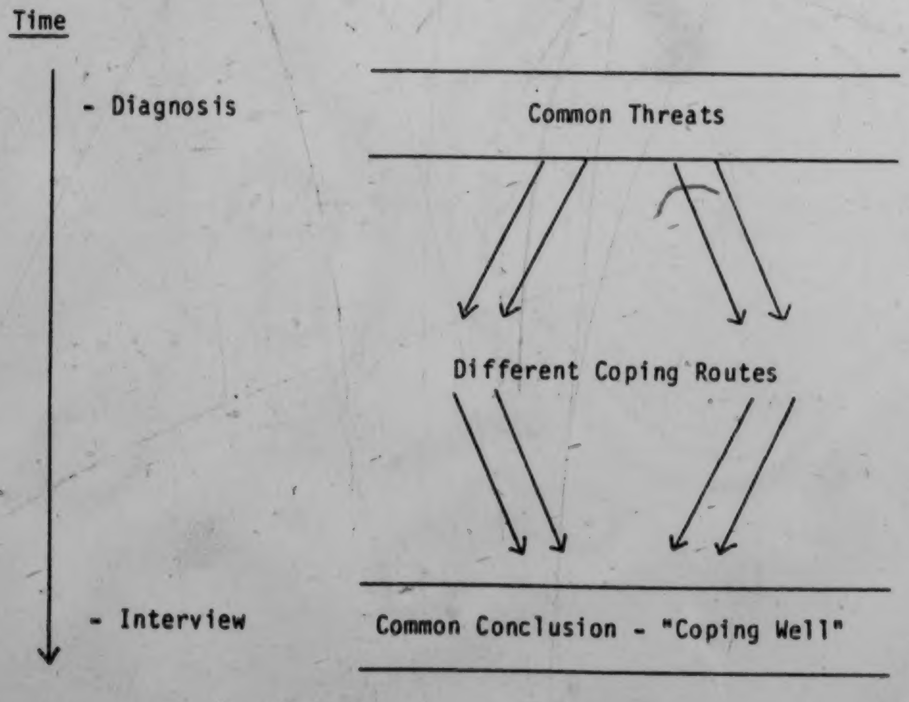
to manage (i.e. master, tolerate, reduce, minimize) environmental and internal demands, and conflicts among them, which tax or exceed a person's resources" (Lazarus and Launier, 1978, p. 311). A depiction of the families' coping efforts is shown in Figure 2. This diagram demonstrates that the families had all encountered similar stressors upon the leukemia diagnosis, and that, many years later, they all had achieved a similar result of self-perceived "good" or "fairly good" adjustment. However, there were some significant differences in the coping strategies used by the various families in travelling between these two points. These variations were directly related to the differences in parental appraisals of the meaning of childhood leukemia for themselves and their families. Parental appraisal and re-appraisal of the leukemia experience is examined in the first section of this chapter. The second section is devoted to sibling appraisal/ re-appraisal of the situation, as well as to a comparison of these perceptions with those of the parents.

Parental Appraisal and Re-appraisal

Many theorists ascribe to the view that coping responses are based upon the personal meaning or definition which an individual ascribes to an event (Hill, 1958; Lazarus, 1966; Lipowski, 1970; McCubbin & Patterson, 1983; Mechanic, 1961; Nerenz & Leventhal, 1983). The most

Figure 2

Simplified Comparison of the Subject Families' Coping


Time
- Diagnosis
Common Threats
Different Coping Routes
- Interview
Common Conclusion - "Coping Well"

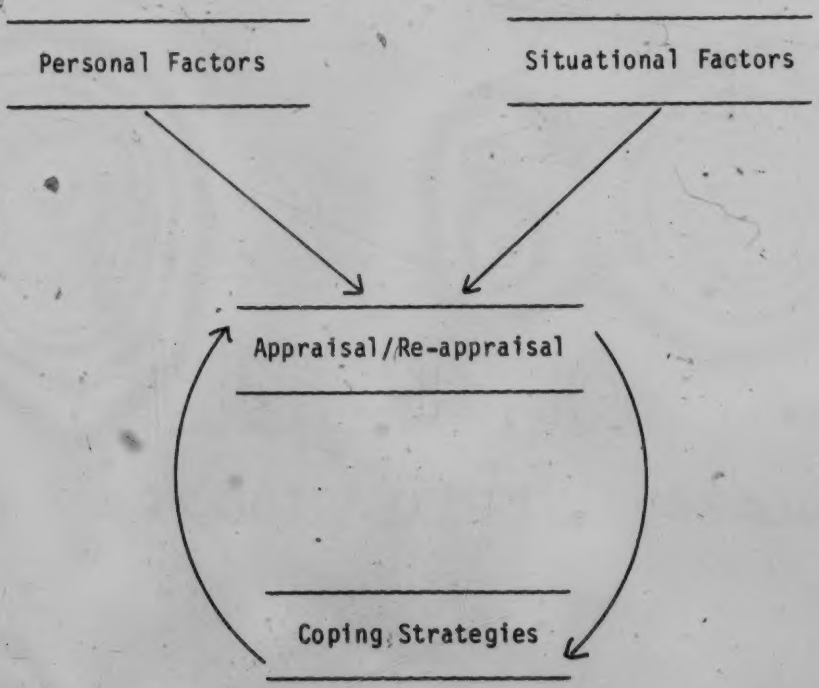
extensively described articulation of this position is the stress, appraisal, and coping framework of Lazarus and Folkman (1984). These authors define appraisal as "a cognitive process that intervenes between the encounter and the reaction (through which)... the person evaluates the significance of what is happening" (p. 52). They identify three types of appraisal- primary, secondary, and re-appraisal. Primary appraisal involves the evaluation of the threat posed by a stressor, and answers the question "What is going on here?" In secondary appraisal, (to be addressed in Chapter 6), the person asks, "What can be done here?". Reappraisal refers to "a changed appraisal based upon new information from the environment" (Lazarus & Folkman, p. 38).

Primary Appraisal- "What is Happening here?"

Lazarus and Folkman's (1984) appraisal/ coping framework (depicted in Figure 3) was used to help label the major factors which influenced parental appraisal. According to these authors, the two (interdependent) sets of factors which affect appraisal are personal factors (especially commitments and beliefs) and situational factors (such as timing, ambiguity, duration, and imminence). Various situational and personal factors which may have influenced family and individual coping with long-term leukemia were described in Chapter 4. However, three specific factors appeared to have exerted

Figure 3

Information adapted from
Lazarus & Folkman's (1984) Appraisal & Coping Model


Personal Factors
Situational Factors
Appraisal/Re-appraisal
Coping Strategies

an especially important impact upon parental appraisal (and subsequent coping). First, the dominant situational factor described by the interviewed parents was the variety of threats to family and ill child well-being which the diagnosis of acute leukemia represented. A second important (personal) factor shared by all the families was their large support networks of friends and relatives. The third main factor influencing parental appraisal was the personal one of perceived ability to control or affect disease outcome.

Situational Factors Affecting Appraisal

Childhood leukemia represented a dual threat for all the families. First, it threatened the survival and well-being of the ill child. At the same time, however, the disease posed a danger to effective family functioning and to the emotional health of all members. Meyerowitz, Heinrich, & Schag (1983, p. 142) state that cancer presents families with a multi-faceted group of stressors, involving "diverse problematic situations".

Initially, the parents were totally pre-occupied with concern for the physical survival of their ill child: "We thought we could lose him at anytime." "As far as we knew, leukemia was fatal, and that's all there was to it." Soon, however, other worries about possible effects of the disease and its treatment on the child began to arise.

For example, one mother stated:

> I remember asking what effect this would have on her ability to have babies. I mean that seemed so silly when we were standing there looking down at this little toddler, but I guess I was thinking about her making it even then.

At this point, worries about the health of other family members also surfaced: "Actually, both of us had the boys checked out, for own our peace of mind, you know." "We got the whole family tested while we were down there, to see what our resistances were... to cancer." Concerns such as these about the health and survival of family members (and about the prevention of therapy- related psychological damage to the ill child) were identified as protection concerns.

Very soon after the initial diagnosis, the interviewed parents began to have concerns about the actual and potential disruption to family life which the leukemia represented, and about the impact of such a disruption upon the children in the family: "I think it was important for us...not to let the illness consume everything." "We have seven of us here. And if we started making changes for one, what would happen to the rest of us?" The parents felt pulled between their own needs and those of the ill child and the well siblings. For example:

> I mean- she needed us there, but we had four other kids at home who needed us, too... It was hard, very hard.
>
> We found we had to make time for the two (other) kids ...we were spending more time in the evenings with them and then back in the morning to spend some time with her. So we were going back and forth...It was hard on us...I got the feeling that I was going a million ways.

The parents also worried about the effect of the leukemic child's new "special" status upon his/ her emotional health: "We didn't want them (the siblings) to resent Mike because he was sick." "We didn't want to make her feel that she was different than the others."

In order to prevent or minimize the damaging effects of the leukemia experience upon family cohesion and upon individual emotional well-being, the parents all felt the need to maintain some sense of normalcy in their families. While all agreed that reducing leukemia-induced family disruption necessitated attempting to "keep things as normal as possible", they disagreed about how "normal" it was possible (or advisable) for their families to be. Concerns of family members about leukemia-related disruptions in family cohesion and in personal happiness and sense of identity within the family were described as normalization concerns.

Personal Factors Affecting Appraisal

The two personal factors which appeared to exert the most influence on parental appraisal were family closeness and the parents' sense of control over the leukemia. As described in Chapter 4, the families were all closely knit, spending much of their time together. Furthermore, the families were similar in their somewhat traditional structures and belief systems. The parents in these families described their children as their first priority: "I've never been able to understand parents where if their child was sick they wouldn't move heaven and earth to get them well again." The families all had ready access to large support networks made up of friends and relatives.

The personal factor which varied among families and which appeared to explain the differences among parental appraisals of the leukemia was the parents' sense of control in the situation. Literature review conducted during the data analysis uncovered that this "sense of control" factor was similar to the "locus of control" concept first postulated by Rotter (1954). According to Rotter, people evidencing an internal locus of control believe that situational outcomes are within their control and are due to their personal efforts and abilities. Those characterized by an external control locus, on the other hand, perceive that event outcome is usually determined by chance or powerful others and therefore is not amenable to personal control. Folkman (1984), Lamontagne (1984), and Brown, Muhlenkamp, Fox, and Osborn (1983) all identify locus of control as one of the most important determinants of coping strategy use. Active coping behaviours (such as vigilance and information-seeking) have been found to be associated with an internal locus of control (Anderson, 1977; George, Scott, Turner, & Gregg, 1980; Lamontagne, 1984), while avoidant behaviours were associated with an external locus of control (Rothbaum, Wolfer, and Visintainer, 1979). These and similar research results will be compared to the study findings at relevant points in the remainder of this paper.

Two of the sets of parents (from the A and B families) could be appropriately described as having an external locus of control because they defined external forces such as the doctor and fate as being largely responsible for the leukemia outcome. The parents from the C and D families used many more references to an internal locus of control, as they described means they had used to affect or control the outcome of their child's illness.

Mr. and Mrs. A described their perceived lack of control in the leukemia situation in the following words:

> If that's what's going to happen, it's going to happen...You can't do nothing about it...We just

> learned how to accept it and then we just hoped and prayed that everything would be okay.

Similarly, Mr. and Mrs. B reported that:

> We really did, we sort of handed her over to him (the doctor). And a lot of the stress...the idea that there was nothing we could do, that was gone then. We didn't know what we were doing, we didn't have a clue. So we said to him, "Just do it."

In contrast to the above parental perceptions, Mrs. D expressed a strong sense of internal locus of control:

> I could no more have sat knowing what I did and left it all in the hands of the doctor than fly in the air. I could never sit back and say, "He's your's-you do what you can." Because they weren't going to miss him like I would. Although the doctors get upset when they lose patients, it's not their kid.

In conjunction with ascribing control over the leukemia outcome to external sources, the A and B parents described the members of the health-care team in glowing, almost omnipotent terms. Mr. and Mrs. A repeatedly referred to the nurses and doctors with whom they dealt as "just fantastic...unbelievable ...good, good people, excellent people" who "looked after us very well". Mr. and Mrs. B felt they received the best possible care for their daughter. "Our feeling was that we had God here in the city, and his disciples out there in (the local hospital)- why did we need to look anywhere else?" Both these families also frequently referred to how "lucky" they had been. They described themselves as: "lucky that it was just for short terms (the hospitalizations)", "lucky that things went as well as they did", "lucky we had have a very supportive family", "lucky we had an honest doctor", "lucky as far between the two of us went" (i.e. good marital relationship), "lucky with his job", "lucky she (the ill child) never got very sick", and "lucky that the other kids were so good". As explained in Folkman (1984), this attribution of much of the situational outcome to luck rather than to personal resourcefulness or hard work is further evidence of the degree to which these parents felt unable to exert control over the leukemia.

In contrast to the A and B families, the C and D parents ascribed noticeably less of their situational outcomes to luck. They also had many more reservations about the abilities of the health-care team (the major external source of control):

> Like sure Dr. X was a good doctor... but there's only so much they can do, because they don't really know what they're dealing with...we didn't know what was going to happen down the road. Was it going to be worth it, like all the suffering and the needles and everything he went through? Was it going to be

worth it?

Mr. and Mrs. C felt they needed to be present during their son's hospital treatments to monitor the care he was given, due to several unpleasant experiences with rough or incompetent health professionals. Mrs. D distrusted the existing health-care system, and suspected that it was dominated by the interests of "the big drug companies...who come up with all these treatments that are so expensive and so toxic, but the people are still dying. And meanwhile, they just keep lining their pockets." At the time of her son's diagnosis, Mrs. D explained that, "There had been enough people around here die of cancer that...we didn't have an awful lot of faith in the system."

Largely consistent with their perceptions of personal control were the sources of early hope for the parents. While the A and B parents received much support from each other, their families, friends, and (with the A's) their church, they described their major source of hope in the initial stages of the disease as "the doctors". For example, these parents stated:

> That was hard the first time (upon initial diagnosis) ...and well, I guess it was about a week later that we were called by Dr. X that he wanted to talk to us... And the way he talked to us, I think that's maybe what got us through.

> We were just basket cases for the first week... (but) they (the nurses and doctors) explained it and tried to make us see that there was hope, and things they could do...and it seemed once we knew what was going on , so we could say, "This is what they're doing today and this is what they are doing tomorrow", then we felt a lot better.

Without a great deal of faith in their doctors, Mr. and Mrs. C described their first year post-diagnosis as, "like what day's it (their child's death) going to be type of deal." The early hope that these parents experienced came from each other and from their belief in God. Similarly, Mrs. D described her child's initial treatment period as terrifying until she and her husband decided to investigate alternate healing methods:

> Once we knew about Laetrile, it made so much sense, we didn't have another sleepless night. From then on we had hope, which we didn't have before. ...Because he was in remission at the time, but we didn't know how long the remission would hold. And he (the doctor) said, "You have to have faith." And I said, "You're a very nice man and that's all very well, but maybe I haven't got enough faith." So that's when we went to the herbal clinic and got the herbs as well.

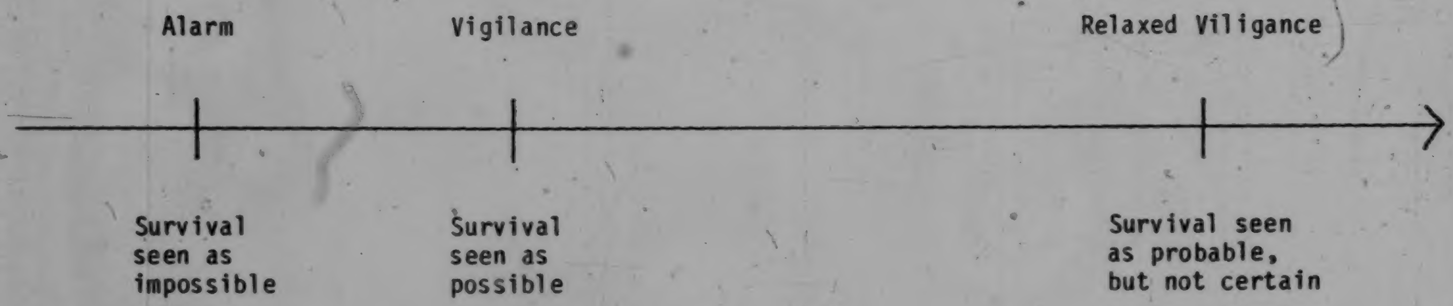
In summary, the parents' primary appraisal ("What is happening here?") after their child's diagnosis of leukemia was influenced by the variety of real and potential threats to individual and family well-being which the disease implied. Awareness of these threats gave rise to protection and normalization concerns about the ill child, the well siblings, and the family as a whole. While the parents had some important beliefs about the family in common, they differed in their perceptions of internal versus external control over the leukemia and its outcome. This difference in attribution of control locus produced significantly different outcomes at the secondary appraisal level ("what can I do about it?") to be discussed in Chapter 6. This finding corresponds with those of Wallston, Maides, and Wallston (1976) and Wolk and Bloom (1978) that demonstrated the existence of a positive relationship between locus of control orientation, situational appraisal, and coping strategy use.

Re-appraisal

During the five to twelve years since the diagnosis, the parents' appraisals of its implications for their ill children and their families had gradually changed. The central aspect of this re-appraisal process was the assessment of the survivor's current health status. Each parent described the stages in his or her own understanding of the survivor's wellness level, and of the events that caused movement from one stage to the next.

Figure 4

Parental Re-appraisal Time-Line


Alarm
Vigilance
Relaxed Viligance
Survival seen as impossible
Survival seen as possible
Survival seen as probable, but not certain

From these descriptions, "appraisal time-lines" were constructed. (Similar time-lines were devised for the well siblings, and are described in the next section of this paper.)

The composite trajectory of the parents' changing evaluations of the status of their ill child's health is depicted in Figure 4. The dominant responses of the parents in this re-appraisal process occurred in three stages: Alarm, Vigilance, and Relaxed Vigilance. The parents' perspectives (normalization versus protection) determined how they translated the emotional responses of Alarm and Vigilance into action, and affected the length of time it took them to reach the Relaxed Vigilance stage.

Alarm

As described earlier, with each long-term survivor the leukemia presented with initially innocuous symptoms which did not disappear as expected. The parents became increasingly uneasy as they took their children to local doctors and were unable to get definitive diagnoses. The ill children were all admitted to local general hospitals for further evaluation, and their parents' apprehension mounted as major intrusive procedures such as bone marrow aspirations were performed. For example,

> When (my wife) phoned me at work and she said, "They're taking him in for a bone marrow", I knew

> what that meant. And that's when I left work and went to the hospital, so I'd be there.

Despite the initial period of building anxiety, the eventual diagnosis of leukemia came as a terrible shock to each of the parents. As illustrated below, the parents at this point all believed leukemia to be a uniformly fatal condition.

> My father looked it up in the dictionary when I first told him and he saw it there, that leukemia is fatal. And he just closed the book and that was it. And that's the way we were understanding it, too.

> We didn't think we'd ever be six (in family size) again...We had thoughts of going in (to the hospital) and never coming out.

> The only thing we knew of leukemia was that you died if you got leukemia...Leukemia was terminal.

The parents all described their emotional responses to this initial evaluation of the situation in similar terms: "We were absolute basket cases." "We were just stunned and in absolute shock." "It was a terrible time- like living a nightmare."

The parents defined the Alarm stage of emotional chaos and belief in a terminal prognosis as lasting from a week to a month after the actual diagnosis. The end to this period was signalled when, after informational meetings with the oncologist, the parents re-appraised their child's situation as one which included the possibility of survival: "It was a totally different perspective after we talked to him (the doctor) because then we understood that it wasn't all terminal. There was hope. There was something they could do." Perceptions of survival possibility varied more according to parental attitude than to information from the doctor. With their faith in the powers of the doctor, the parents in the A and B families interpreted the prognoses they were given as relatively good news. For example, the A's stated, "He told us that she had a 50% chance...her odds for survival were good." As evidenced below, however, the C's and D's interpreted similar prognoses as threatening:

> It was only after a year that we could really believe the 50 part that said, "Okay, he's going to live." Because like a year previous we didn't have the year, we didn't have the month, you know...Like, 50/50 doesn't seem like a lot when that's your child.

> (The doctor told us)...45% were doing well... and I thought that sounded pretty good until we got in the car and I realized 55% were not doing well- that scared me.

This arbitrary designation by parents of similar prognostic statistics as either "good news" or "bad news" was also noted by Cohen (1986). She found that parents of children with cancer were unable to deal with the subtleties of survival rate percentages, and tended to convert that information into a binary choice between "good" or "bad" news. The particular attributions her sample of parents chose were determined by their psychological make-ups, rather then by the specific odds they have been given.

Vigilance and Relaxed Vigilance

Vigilance is the re-appraisal stage which the parents entered when they decided that their child's survival was possible. Relaxed Vigilance did not occur until years later, when the parents finally came to believe that their child's survival was probable (although not certain). The parents could all be appropriately described as being in the stage of Relaxed Vigilance at the time of the interviews.

After entering the stage of Vigilance following the acceptance of the possibility of survival, the parents quickly became aware of the multi-faceted threats (in addition to death of the ill child) that the leukemia represented. It was at this stage that a "balancing the demands" decision between normalization and protection orientations was made. While this decision led to differences in the way the parents acted out their vigilant awareness, the anxiety resulting from living with the constant uncertainty of childhood leukemia remained the dominant emotional theme:

> I'd often looked at Mike and wondered how long we could keep him...It was hard. I thought about it all the time.
>
> For the first five years I was totally neurotic about the (laetrile) diet and everything...It was a lot of hard work, but the thought that we might lose him drove me on.
>
> Yeah, for years after, every cold, every bruise, I'd think, "Is this it? Is this it back again?"

As the length of the remission increased and the milestones of therapy (i.e. end of chemotherapy, end of intrusive procedures, etc.) were passed without event, the parents' anxiety and need for vigilance slowly decreased: "The better things were going, the more we could relax." "I guess the longer the time between visits, the better it seemed. Like we don't have to go so often, so he must be doing better." When asked when they had made the transition from assuming "possible survival" to "probable survival", some parents described specific events which to them marked turning points in the child's treatment. For example:

> I think it was when her visits went down to once every six weeks. I decided that if she could go that

> long without any problem, we probably had 'er beat.
>
> Well, when they stopped her chemotherapy, that was a bit scary, you know. But I held my breath and when she was fine and she stayed okay, I thought, "She's probably going to make it."

For some parents, the transition to Relaxed Vigilance had not been of their own active choosing, but instead had taken place as a gradual evolution. However, these parents were able to identify behaviours or feelings that indicated that they had, at some point, made the shift in thinking away from the earlier assumptions of the Vigilance phase. For example:

> I thought it was the earmark of graduating or something when I totally forgot her latest appointment. They had to phone me at work and they said, "Aren't you bringing her in?"...I think that's a real fundamental change, and I think it only happened in the last couple of years, as well.
>
> When he was younger, to scold him or anything we used to find it very, very difficult...whereas now we think, well, if he doesn't get this idea now, he's going to run into trouble ten years down the road....We are preparing him for life now.

Even with the more positive outlook associated with Relaxed Vigilance, however, the parents all remained acutely aware of the continuing uncertainty about the long-term prognosis for "cured" acute lymphoblastic leukemia. They all made statements similar to Mr. B's that "it tends to stick in your mind...that fear that it might come back." This awareness made all of the parents reluctant to use the word "cured" to describe their children. They expressed reservations such as:

> I never use the word cured. Never...We sort of have a taboo with it...It's like a jinx...You're never cured. It's always you're in remission or off-treatment or whatever. But never cured.

> I don't think you can use the word cured because it's something they'll always have- somewhere...It's like an allergy- they'll always have a particular sensitivity for the rest of their lives...And anyways, I'm superstitious about using the word "cured". It's like tempting the fates.

Therefore, while the parents were willing to relax their vigilance and assume probable instead of just possible survival, they were unwilling to believe in certain survival and therefore to discard their vigilance over their leukemic children all together.

Sibling Appraisal and Re-appraisal

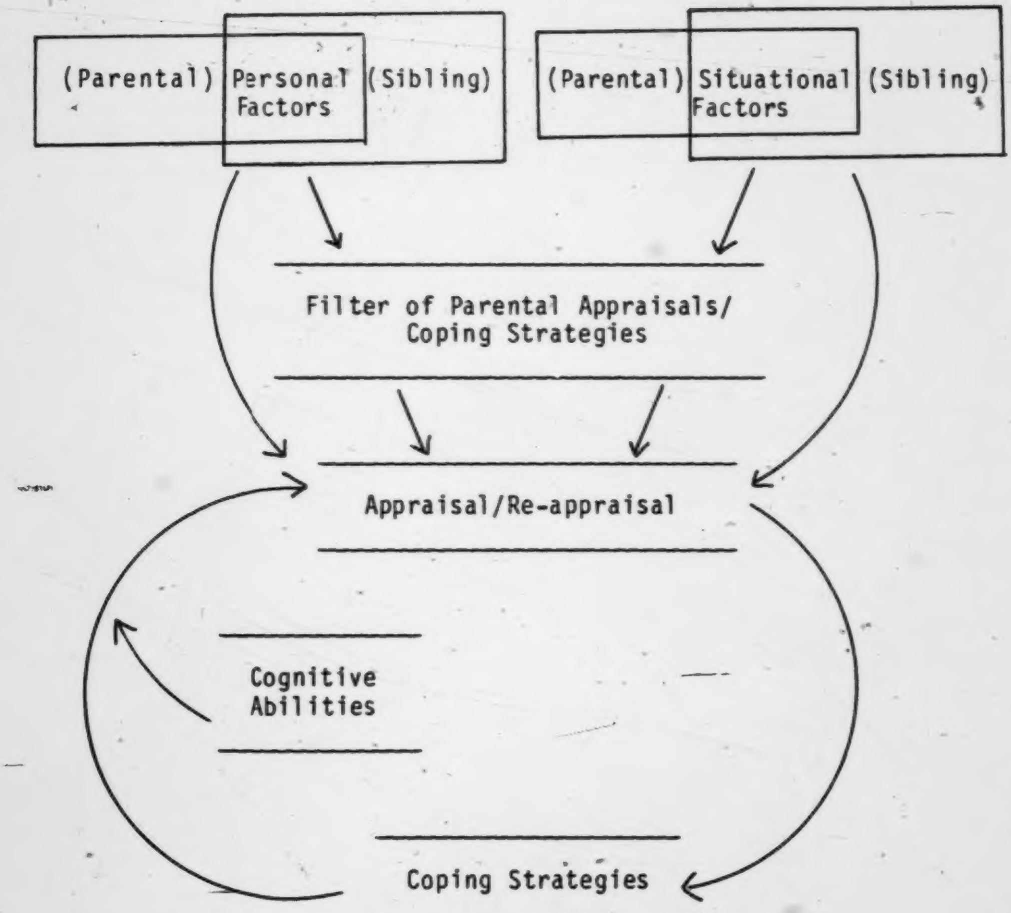
As with their parents, sibling coping strategies used in living with long-term childhood leukemia were determined by their appraisals of the meaning of the situation for themselves, their families, and their ill brothers or sisters. However, there was an important difference between the appraisal/ coping strategy relationships evidenced by the parents under study and those which were described by their children. While the coping process for the parents could be appropriately represented with the use of Lazarus and Folkman's (1984) framework (Figure 3), this diagram must be altered somewhat if it is to accurately reflect sibling coping. Figure 5 presents the Lazarus and Folkman model with modifications appropriate for the appraisal/ coping relationships evidenced by the well siblings. This figure shows that sibling appraisals and re-appraisals were significantly affected by parental appraisals and coping strategies, and by the siblings' immature (but evolving) cognitive abilities.

Primary Appraisal- "What is Happening Here?"

As depicted in Figure 5, the situational and personal factors which mediated the siblings' primary appraisals overlapped significantly with those of their parents. The most notable characteristic of sibling appraisal, however,

Figure 5

Appraisal/Coping Model for Siblings


(Parental)
Personal Factors
(Sibling)
(Parental)
Situational Factors
(Sibling)
Filter of Parental Appraisals/ Coping Strategies
Appraisal/Re-appraisal
Cognitive Abilities
Coping Strategies

was the filtering impact of parental appraisals and coping strategies upon the leukemia-related information which formed the basis of the siblings' interpretations. Parents controlled access to much of this information: "They brought this book home about leukemia but we weren't allowed to look at it by ourselves until we were in grade 5, in case we got too upset." When disease-related information was made available to the siblings, parental appraisals provided the children with ready-made explanatory frameworks for use in its interpretation. One set of parents, for example, couched their explanations in the following terms:

> We told them that it was dangerous enough, that cancer was dangerous... but we just said we'll hope and pray for the best. You know, we've got a good doctor and I'm sure he'll find it. And she'll be fine.

Another mother described her explanations in terms of brainwashing:

> We brainwashed (them) that carrot juice killed the cancer cells and Laetrile killed the cancer cells, so that every time he (the ill child) took a pill or drank juice, he was killing the cancer cells...I think us having that positive attitude- it rubbed off.

Given their lack of access to outside information and the persuasive power of their parents' interpretations, it is not surprising that the siblings' appraisals of their leukemic brother or sisters' long-term prognoses echoed those of their parents. In the A and B families, where the parents had assumed an external locus of control over the disease and its outcome, the siblings agreed that little or nothing (except medical therapies) could be done to control the disease or prevent relapse. On the other hand, children from the more protection-oriented C and D families believed that long-term outcomes for their siblings would be good, as long as protective measures such as vigilance and dietary restrictions were maintained. According to these siblings:

> If he eats healthy, I guess, he can be safe. But if he starts bad habits like smoking or drinking or eating the bad stuff, I guess that will leave an open gap (in his immune system) for it to set in again.

Because they were largely removed from the "scenes of the action" (i.e. the hospital, the consultations with the doctor, the clinic visits), the siblings were dependent upon their parents for information about leukemia and its implications, and the current status of their ill brothers or sisters. While some of this information was explicitly conveyed by the parents, the siblings relied heavily upon inference and deduction in their appraisals of the situation. This reliance upon inferred and indirect information had two main roots: the parents' reluctance to talk about the disease and the siblings' immature cognitive abilities.

As described in Chapter 6, "not talking about it" was a key parental coping strategy in dealing with the long-term uncertainty of childhood leukemia. In order to protect themselves and their children from undue anxiety, and to maintain the atmosphere of family "normalcy", parents provided only limited disease-related information to their children. As one parent explained:

> We never thought of hiding it from them. But why talk to them about it, unless they say they want to talk about it? And I guess that's more or less the approach we took.

Only one set of parents stated that they actively tried to up- date their explanations about the disease as their children grew up: "We gradually increased it into them...It was pretty well, as we learned it, we explained it to them." In the other families, the siblings were typically given little information to supplement the often simplistic explanations they had been told at diagnosis.

Because many of the siblings had been very young at the time of initial diagnosis, the general lack of follow-up information meant that some of the younger siblings had never had the disease explained to them by their parents. Instead, they based their understanding upon bits and pieces of information they had picked up from a variety of sources. For example, when asked for a definition of leukemia, one of the younger siblings stated:

> I know it's a cancer...Cancer is a serious disease ...No, nobody told me that. I just figured it out for myself...from sometimes when I hear my mom talking about it on the phone.

A survivor described the efforts of her youngest sister, age three at the time of the diagnosis, to gain information:

> Like she used to come up to me sometimes and ask how I was. And I'd say. "I'm fine, I'm fine." She'd ask if I have cancer, if I had cancer, and I's say, "Yeah, I had blood cancer."...She was probably hearing a bit of the stuff around the house. Maybe no-one knew what to tell her. So then she'd come to me and ask, "Are you okay?" And, "What did you really have?" And, "Are you going to die?" And, "Are you okay?" And I'd go, "Yeah, I'm okay, I'm okay."

While the siblings occasionally used information from biology teachers, library books, television shows, and newspaper articles to increase their understanding of leukemia and its implications, their general tendency was to avoid actively seeking outside information. As described in Chapter 6, the dominant sibling coping strategy was "not talking about it", which included "not thinking about it" and the "limited seeking of information". These strategies were designed to decrease the anxiety related to the life-threatening diagnosis and uncertain prognosis. As one sibling suggested, "Maybe we don't ask that many questions about it because we don't really want to find out the truth."

As well as having limited access to information, the younger siblings also had difficulty making use of that information which was available. Displaying the cognitive limitations of their age group, the pre-teen siblings relied more upon evidence they could see or hear in making their appraisals than upon the sometimes vague, confusing, and incomplete explanations from others. For example, when asked what had convinced them that leukemia was a serious disease, the siblings listed the following seven factors:

1. When the ill child lost his or her hair;

2. When siblings were not allowed to visit the ill child in the hospital;

3. When the ill child had to endure numerous painful or unpleasant therapeutic measures;

4. When the ill child looked seriously physically ill;

5. When numerous measures to protect the ill child were instituted;

6. When parents said the ill child could die;

7. When parents were upset and not talking about what was wrong.

Of the seven factors listed above, it is notable that only one (#6) pertained to information explicitly conveyed by the parents. By far the majority of the evidence which had convinced the siblings of the life-threatening nature of the leukemia was of an indirect and experiential nature. For example, the only factor mentioned by every sibling was their brother or sister's baldness (#1):

> When he got home, his hair was falling out. And that's when I figured it was quite serious. I thought he'd die or something. Like, I was so young I thought he was falling apart.

The children were worried primarily by evidence of the ill child's suffering (#3 and #4), by the unexplained (#1, #2, and #7), and by changes in routine (#5):

> When my mom went and locked herself in her room... and wouldn't talk to me...I worried...because if the whole family is all crying and stuff, it's got to be something wrong.

> Probably when we started going to the hospitals a lot. Going for all those different things, the tests and everything. That's got to be something serious then- not the normal cold.

Even when siblings were given information, they tended to trust what they experienced more than what they were told:

> Well, they told me he was going to be okay, but I didn't believe them because they acted so worried all the time. So I worried, too.

In summary, sibling appraisal (and its relationship to subsequent coping) was less straight-forward to assess than the Lazarus and Folkman (1984) framework depicted in Figure 3 would suggest. This framework was devised to explain adult coping, and does not address two factors which powerfully influence the appraisal and coping of children within the context of their families. These factors are the children's immature (but evolving) cognitive abilities, and their reliance upon their parents for information and explanations. Combined with the "not talking about it" strategy employed by all family members, these factors contributed to the siblings' reliance upon indirect and inferred information in making their situational appraisals- and to their minimal understanding of leukemia and its implications.

Re-appraisal

The combined impact of limited direct information and predominantly inferential reasoning from concrete evidence rendered the sibling appraisal time-lines significantly different from those of their parents. These two factors also contributed to the fact that the sibling re-appraisal time-lines showed two distinct variations. Figure 6 depicts a cumulative time-line of the siblings' changing appraisals of the health status of their ill brothers or sisters. Figure 7 demonstrates the two main variations in this time-line. In both figures, the four stages of the siblings' re-appraisal of the meaning and implications of the leukemia are shown: Awareness, Alarm, Vigilance, and Relaxed Vigilance. Sibling progress through these four stages was powerfully affected by certain parental coping strategies, and by the increasing sophistication of sibling cognitive abilities. It was also influenced by the siblings' tendency to assume wellness unless presented with evidence to the contrary.

Awareness

During Awareness, the siblings were aware that "something" was wrong with their leukemic brothers or sisters, but did not perceive the seriousness of the situation. For siblings old enough to have noticed and remembered the events surrounding the diagnosis of the

Figure 6

Cumulative Sibling Re-appraisal Time-Line

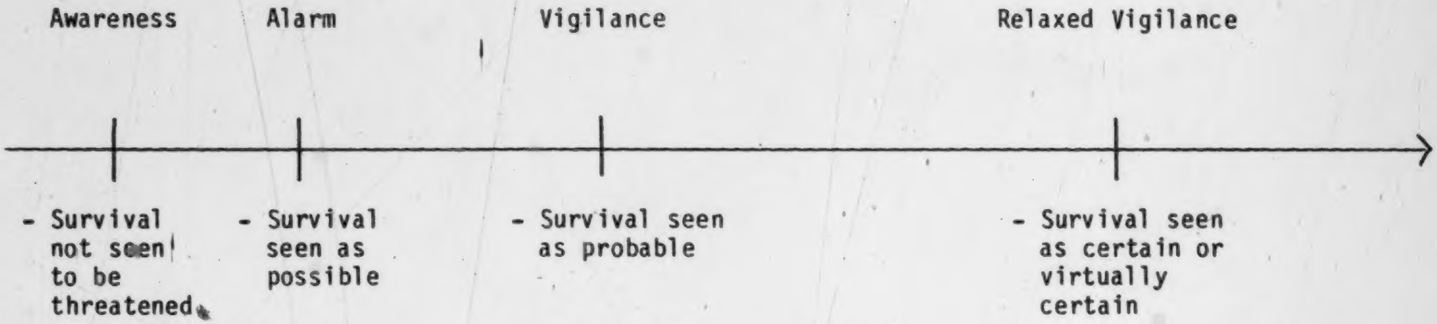
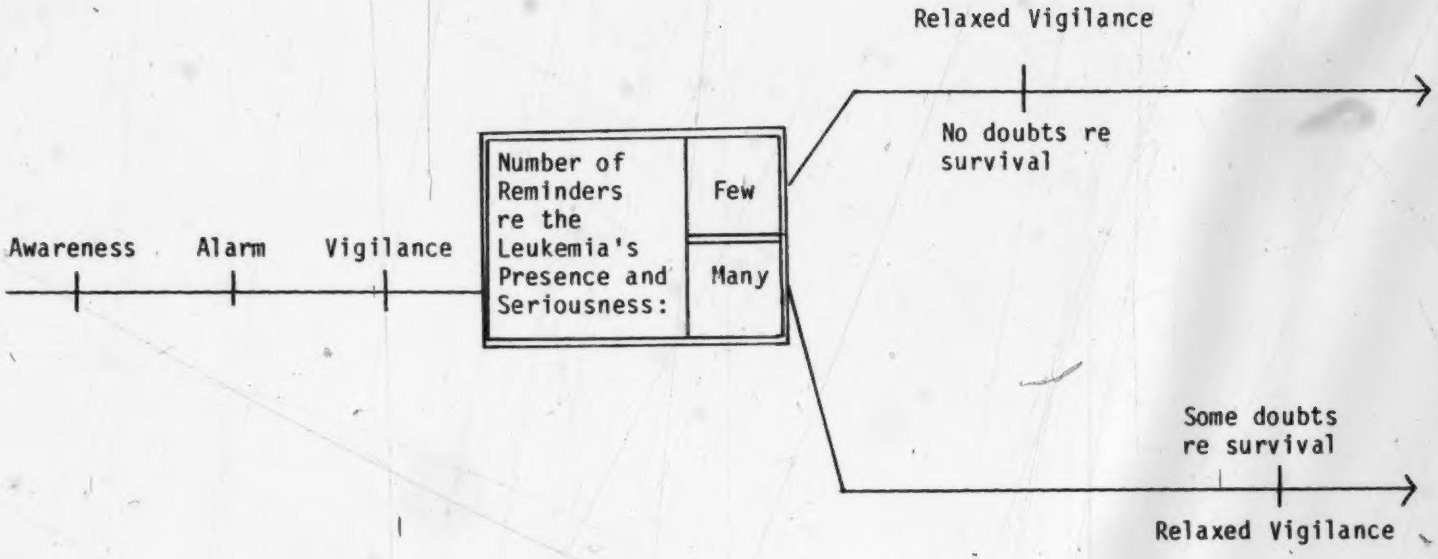

Awareness
Alarm
Vigilance
Relaxed Vigilance
- Survival not seen to be threatened
- Survival seen as possible
- Survival seen as probable
- Survival seen as certain or virtually certain

Figure 7

Two Variations of Sibling Re-appraisal Time-Line


Awareness
Alarm
Vigilance
Number of Reminders re the Leukemia's Presence and Seriousness:
Few
Many
Relaxed Vigilance
No doubts re survival
Some doubts re survival
Relaxed Vigilance

leukemia, initial awareness of "sickness" followed a gradually unfolding pattern similar (although less intense) to that of their parents'. Early mild symptoms in the ill child were either ignored or dismissed as unimportant: "I didn't know why they were taking him to the hospital, cause I thought it was just a case of the flu." More exaggerated symptoms and hospital admissions for diagnostic evaluations did cause some concern for the older siblings. Overall, however, the children tended to assume that the situation was only a minor change from usual routines and was not a cause for alarm. As one explained:

> At first, I didn't think much of it. I thought they must be doing all they can to, well, make sure he's okay. So, I just assumed that everything's okey-dokey.
>
> I just thought there wasn't that much going on. So I didn't feel bad cause I thought it was just like the measles or something, except a badder case.

The siblings' tendency to assume the leukemic child's wellness unless faced with concrete evidence of illness emerged as one of the dominant characteristics of their re-appraisal styles. It contrasted with their parents' inclinations towards worry and fear of the unknown in the face of uncertain diagnoses and prognoses.

Alarm

Siblings entered the Alarm stage of re-appraisal when they learned the seriousness of the diagnosis, and feared the ill child's death to be possible- and perhaps probable. The siblings were not informed of the leukemia diagnosis until several days to a month after their parents were first told of it. By that time, however, several of the older siblings had already noted enough parental anxiety to seriously concern them: "I guess I...noticed by their expressions...The main way I found out by was how they acted." Other siblings became alarmed only after being told of the name and nature of the illness. For example:

> Then she said it was a cancer...That scared me, because you know cancer is a fatal disease.
>
> My Mom was really worried and she sat us down and she brought out the book. And she said, "This is what she's got." And it was, you know, it didn't hit me until she showed me that. And I said, "Oh, holy mackerel!" You know, it was kind of scary at first.

The early Alarm responses described by the siblings were fear for their ill brothers or sisters and confusion about what to expect. To deal with such disruptive emotions, the siblings initially sought information and reassurance from their parents: "Mainly we just kept asking them, "Is he going to be alright? And what is it? Like, what is leukemia?" In most cases, reassurance from parental answers and from the return home of the relatively healthy-appearing leukemic child caused the siblings to move quickly from their Alarm appraisal of probable death to a more positive evaluation of the situation. Examples of such reassurance include the following:

> They told us that they had all this good technology nowadays that would make him better.
>
> Well, it was a little tense when she first got back home. But pretty soon she was playing again just like before, and we started bugging her, and things felt like they were going back to normal.

Vigilance

During Vigilance, the siblings appraised their leukemic brother or sister's prognosis as probable survival, but remained well aware of the possibility of death. The two main sibling responses during this stage were worry and the perceived need to "keep an eye on" the ill child. The duration of Vigilant re- appraisal varied greatly, from two weeks to four years, with most of the siblings fitting in near one extreme or the other. Duration times were determined by the amount of reminders and negative information about the disease to which the siblings were exposed. The two major patterns of transition from Vigilance to Relaxed Vigilance are depicted in Figure 7.

The upper appraisal time-line in Figure 7 depicts the response of siblings who had very few reminders of the leukemia's presence or nature, while the lower line represents the re-appraisal process of siblings receiving frequent and/ or powerful reminders. These reminders typically were in the form of changes in family routine designed to "protect" the ill child and the witnessing of traumatic diagnostic or treatment procedures. The major differences between the re-appraisal processes represented by the two lines in Figure 7 were in the amount of time the children spent in the worried stage of Vigilance, and in their degree of certainty about long-term survival once in Relaxed Vigilance. These two factors were related, in that the longer the sibling had spent in Vigilance, the less certain they appeared to feel about the leukemic child's "cure" once they were in Relaxed Vigilance.

Due to their tendencies to both assume wellness and rely upon inference for information, the siblings quickly revised their appraisals of the leukemic child's state of health in a positive direction unless they continued to be exposed to negative indicators and reminders. For example, in the A family, the disease was rarely mentioned and the siblings did not attend clinic visits with their sister. As noted by Mrs. A: "It really didn't affect the rest of the family. They really had no awareness of what was going on at the clinic." Also, in this family the leukemic child remained generally well and few measures to "protect" her were introduced by the parents. After worrying and/ or watching her for a few weeks post-diagnosis and seeing no new evidence of threat, her siblings assumed that "everything was back to normal". They made statements such as:

> Like when she was running, when she was playing, it seemed that she wasn't going to die then because she was just happy and herself.
>
> Because if she's looking so well, and responding so well, then you never really think about it....Honestly, it didn't even seem like she was sick, you know.

By only a few months after diagnosis, the A siblings had revised their appraisals of the status of their leukemic sister to "basically cured". They therefore fit into the upper appraisal line depicted in Figure 7. As one brother stated, "I thought it (the leukemia) had passed, you know, in a few, maybe two or three weeks." It was not until years later that the A siblings became fully aware of the long-term uncertainty which the diagnosis of acute childhood leukemia implies: "Actually, I only figured out how serious it was about a couple of years ago (at age nineteen or twenty)." "I didn't know how bad it was until I was about sixteen or seventeen, and it really hit then that, you know, it is fatal."

Children who described themselves as staying in the Vigilant stage of re-appraisal for prolonged periods (the lower time-line in Figure 7) were those who had continued to encounter distressing information about their siblings' leukemia long after the diagnosis. Such information was mainly in the form of witnessed painful therapeutic procedures and leukemia-related activity restrictions. Subjects who had accompanied their ill siblings to their clinic visits and had listened to their screams (during bone marrow aspirations and lumbar punctures) remained especially frightened of the disease and its treatment. As described by these children:

> I'd hear her screaming and crying and oh, it just drove me crazy, you know. Like what are they doing to my sister in there?...It drives me crazy just thinking about it.

> He'd be in there getting shots in the back and you could hear him just screaming he felt so bad. You didn't even want to stay around there...Cause like I figured he'd die or something. Cause it sounded like they were really hurting him on purpose, with the screaming and stuff.

One factor which contributed to the siblings' anxiety over painful invasive procedures was the "not talking about it" coping strategies used by all family members. Fear sometimes caused the siblings to withdraw into themselves rather than seek reassurance or information:

> Like I was just so worried that I couldn't think of what to ask them. And I thought that they'd be so worried, too, that they couldn't answer.

In other cases, reassurance was occasionally not available when it was sought:

> Nobody in there (the clinic play-room) to talk to that I knew...They'd (the parents) usually be talking to the nurse or something, so when I tried to interrupt the conversation, they'd just shoo me away and say, "Go play".

Significantly, these siblings identified the point at which their ill brother or sister became "cured" or "better" as the point at which the major intrusive procedures were discontinued: "It was when she stopped having bone marrows, like when they stopped poking needles into her back and things like that. That's when I mostly thought she was cured." When the disturbing evidence stopped, the siblings assumed that the situation was no longer serious: "I felt it meant if you didn't have to take (the spinal taps) anymore, than you don't have to worry about it coming back or it getting worse".

Besides awareness of painful procedures, the major "evidence" that caused siblings to continue to appraise the leukemia as actively threatening was the presence of special protective rules and restrictions in their families. Having to adjust their activities (and those of their family) to "protect" one child served as an ongoing reminder for the siblings that the leukemic child was "sick", "might catch something", and "still had to be careful". One sibling from the C family described this awareness in the following terms:

> Well, we got to play with our friends a lot, and when I saw him in there and asked my mom, "Why can't he come out?", she said, "Mike really can't come out." And so I guess...I saw how unhappy he was and was always asking my mom, "Why can't he go out?" and "Is he really okay?"

The worry over the implications of the C family's protective regulations expressed in the above quote was not shared by siblings of the D family where even more extensive protective rules were in force. This difference could probably be best attributed to variations in the way the two sets of parents explained their protection measures to their children. The D's protective strategies involved significant life-style changes for the family, but were presented as potentially curative for the ill child. The C's protective measures, on the other hand, were explained as stop-gap preventative rather than curative strategies, and as such did not provide their children with the reassurance of "having an answer" for the disease. Instead, these measures served to repeatedly remind the C siblings that something dangerous- and largely uncontrollable- was affecting their brother.

Relaxed Vigilance

The fourth stage of the sibling appraisal time-line (Relaxed Vigilance) began at the point that the children felt relatively certain about their ill sibling's long-term survival. The degree of certainty about "cure" was affected by the amount of time the well sibling had spent in Vigilance, and by the child's level of cognitive understanding. New doubts and questions often arose as the siblings gained the abstract and hypothetical reasoning abilities associated with the onset of adolescence.

As depicted in Figure 7, the children who had little exposure to negative information during Vigilance and who had subsequently assumed very early on that their ill sibling was "cured" (or at least out of danger) tended to have few concerns in Relaxed Vigilance about the survivor's long-term prognosis. For example, siblings from the A family felt that: "She's cured ...It's all gone...she's alright now." "I think it's all over because, well, it's over. Cause nothing happened...She looks just fine to me." On the other hand, the interviewed siblings who had been exposed to ongoing evidence of the serious nature of the ill child's disease began to infer "cure" only when the negative information (especially the painful procedures) stopped. Even then, however, they continued to have some disturbing lingering doubts about the long-term prognosis. It appeared, therefore, that prolonged earlier periods of vigilant concern about possible sibling death left residues of apprehension which, while decreasing over time, continued to surface occasionally to haunt the children.

Those children who had been the slowest to enter the appraisal stage of Relaxed Vigilance, then, were also least certain about the assumption of "certain" or "almost-certain" survival of their leukemic sibling. Overall, however, the siblings appeared to be considerably more convinced of the likelihood of a positive outcome to the situation than were their parents. This view may be a further indication of the siblings' tendency to assume wellness unless shown otherwise.

Of interest was the finding that the siblings' faith in a positive disease outcome was disturbed by new perspectives and anxieties which spontaneously arose between their tenth and fourteenth years of age. This age-period marks the onset of formal operational cognitive abilities in the child, which involve the development of aptitudes for abstract and hypothetical reasoning (Phillips, 1975). The development of such abilities would enable the child to consider new aspects of the leukemia situation, and to ask more penetrating questions about its implications. It is therefore not surprising that most of the siblings identified this age period as an important re-appraisal point along their awareness trajectories. For example:

> When I first understood what leukemia was? Well, maybe about twelve. Twelve or ten. I started asking my sister, then, too, what had happened.

> Since I was about ten or around that age, I started asking more questions...so now I'm beginning to understand more.

For most of the siblings, the onset of formal operational cognitive abilities occurred during Relaxed Vigilance and caused them to re-assess their earlier assumptions of certain or almost certain cure. Some siblings simply put these worrisome thoughts aside: "There's no use thinking about it, because she's going to be fine." Others found that time and a certain amount of mental agonizing were required before their concerns could be worked into a less stress-producing overall perspective. As one explained:

> I used to worry about it quite a bit...(and) I know

> there's still a small chance it could come back. A small chance. But I think he's doing okay, and they're keeping a good eye on him. So now I'm about 99% sure he'll be alright.

In summary, the sibling re-appraisal time-lines were strongly influenced by parental coping responses to the leukemia and by the level of the child's reasoning abilities. They also were affected by the sibling tendency to assume wellness unless presented with evidence to the contrary. Those siblings who moved most quickly through the four re-appraisal stages, and who felt most positive about the ill child's long-term prognosis, were those who had been exposed to the fewest worrisome reminders about the disease. Some prognostic doubts and concerns typically arose with the development of more sophisticated intellectual skills in early adolescence.

Comparison of Parental and Sibling Re-appraisal Time-lines

In comparing the sibling and parental re-appraisal time-lines of the health status of the leukemia survivor, several major points are worth highlighting. These points pertain to differences in perceptions about disease prognosis, length of time spent in Vigilance, and variations among re-appraisal processes.

The siblings' early awareness of "sickness" in the ill child was noticeably less anxiety-tinged than that of their parents', due to the siblings' lack of detailed information and their general assumption of wellness. The onset of the Alarm response in the siblings occurred days to weeks after their parents entered the Alarm stage, and usually did not begin until the nature and implications of leukemia were explained to them. While the parents viewed death from leukemia as inevitable during Alarm, their children interpreted it to be possible, and worried that it might be probable.

The parents entered the Vigilance stage of appraisal after being convinced of the possibility of leukemia survival by their doctor. The siblings entered the same phase after information from their parents and the evidence of a relatively healthy-appearing leukemic sibling showed them that the disease was being treated and that survival was probable. Siblings, especially those with little or no exposure to negative or frightening information about the disease, remained in the stage of Vigilance for a much shorter period than did their parents. Once in Relaxed Vigilance, the parents also remained much more cautious than the siblings about the survivor's long-term prognosis.

Parental re-appraisal time-lines showed more similarities among themselves than did those of the siblings. There appear to be two reasons why the sibling time-lines varied so much more than the parents'. First, the parents received much of their information about the disease from the same source (the health- care team), whereas the siblings were dependent upon the differing interpretations of the situation presented by their parents. Second, because the siblings ranged in age from two to thirteen years at initial diagnosis, the subsequent onset of concrete and formal operational cognitive abilities was somewhat variable in its timing and impact upon sibling re-appraisal.

Summary

In summary, situational appraisal was found to be the major determinant of subsequent coping strategy use. For both parents and siblings, this appraisal included primary appraisal ("What's going on here?"), and ongoing re-appraisal. The main situational factor affecting parental appraisal was the multitude of possible leukemia-related normalization and protection concerns. The most important personal factor which influenced parental appraisal was their perception of personal versus external agent control over the disease outcome. Parental re-appraisals of their leukemic child's prognosis moved through three stages- Alarm, Vigilance, and Relaxed Vigilance. The parents progressed from perceiving their child's death from leukemia as certain, to believing that survival was possible and then probable.

Sibling appraisal was strongly influenced by parental appraisals and coping strategies, and by the level of the sibling's cognitive abilities. Sibling re-appraisal of the ill child's status included four stages- Awareness, Alarm, Vigilance, and Relaxed Vigilance, and reflected a tendency to assume the ill child's wellness unless presented with evidence to the contrary. Two main variations in sibling re-appraisal time-lines were noted, resulting from the differing amounts of negative information about the disease to which the siblings were exposed. Sibling and parental re- appraisal time-lines differed in several ways, including their degree of variability, the length of time spent in Vigilance, and the long-term certainty about survival. In both cases, however, these appraisals were intimately intertwined with the coping strategies and long-term outcomes described in Chapter 6.

Chapter Six

Coping Strategies and Consequences: Findings and Discussion

This chapter focuses upon the coping responses of the interviewed families to living with long-term leukemia in a child, based upon their situational appraisals as discussed in Chapter 5. In the first section of this chapter, the parental responses to the secondary appraisal question of "What can be done here?" will be examined. Specifically, the decisions the parents made on how best to balance their efforts to meet the demands represented by their normalization and protection concerns will be presented and discussed. The coping strategies which complemented each balancing-the-demands decision will then be described. The use of specific coping strategies will be compared and contrasted among families and between parents and well siblings. Apparent long-term consequences of these coping styles for the well siblings and their families will be discussed. Hypothesized responses and consequences for families showing more extreme imbalance than any of the subject families in their normalization versus protection orientations will also be suggested.

Balancing the Demands

The parents' primary appraisal of the situation at the time of their children's diagnoses represented an acknowledgement of the threat the disease presented to the existence and well-being of both the ill children and their families. Awareness of this dual threat caused two sometimes competing sets of concerns to arise in the parents' minds- those related to protecting the ill child from relapse and death and those related to "normalizing" the experience for all family members. In secondary appraisal, the parents were faced by the difficult question of "What can be done to cope with these threats?". While all the parents acknowledged the needs represented by both their normalization and protection concerns, they tended to focus upon meeting the demands of one over the other. This emphasis was affected by their primary appraisal of the situation, and in particular was determined by their locus of control over the disease outcome. As described in the following section, those parents with an external locus of control concentrated upon normalization rather than protection concerns and strategies. The parents with an internal locus of control emphasized protection over normalization issues.

The parents' "balancing the demands" decisions could result in orientations ranging from total emphases upon either normalization or protection issues, to any degree of balance between the two. The points of balance chosen by each of the four subject families are depicted in Figure 8. These normalization versus protection orientations were determined by the parents in the first few weeks after diagnosis, and remained consistent throughout the ensuing years. They were crucial determinants of the coping strategies used by each of the subject families, which are discussed in the following section.

Coping Strategies

In this section, the coping strategies used by the parents and siblings to deal with long-term acute leukemia in a child will be discussed. First, those strategies used by the normalization-focussed A and B parents will be presented. Second, coping strategies of the protection-oriented C and D parents will be described. Third, parental coping strategies used by both "normalizers" and "protectors" will be discussed. Finally, strategies used by the siblings will be described, and compared with those used by their parents.

The "Normalizers"

The A and B parents' external locus of control led them to believe that there was little they could do to protect their leukemic child from relapse or disease complications. Instead, they placed their faith in the doctor to look after most of their protection concerns. These parents then concentrated upon coping strategies that dealt with their normalization concerns.

According to Krulik (1980), Robinson (1985), and Klein (1976), normalization is "those strategies employed by families with ill or handicapped members to live as normal a life as possible". In addition, Wolfensberger (1972), Anderson (1981), and Kling (1980), note that normalization is an ideology which reflects the positive social value our society attributes to "being normal". In the present study, normalization is defined as those efforts instituted by family members to minimize both leukemia-related change within the family and differences between the ill child and the well siblings. This definition alludes to the two aspects of normalization described by members of all the subject families. According to these individuals, keeping things "normal" within the family after the diagnosis of leukemia included both "keeping things the same" and "maintaining equality amongst the siblings". Various facets of these two components are described below.

Futterman and Hoffman (1973) describe normalization as involving "adherence to familiar routines (and)...continuation of usual patterns". Believing that their ill child's physical health was in the hands of the health-care team, the A and B parents concentrated upon maintaining the psychological well-being and smooth functioning of the family unit by "keeping things the same": "We did as little changing as we could, only what we had to. We tried to keep it as normal as we could." Both Mr. and Mrs. A and Mr. and Mrs. B saw the introduction of leukemia-related change into their established routines as representing a threat to the harmonious and effective interaction of their families. These parents defined their families as functioning well before the diagnosis, and assumed that significant alteration of pre-established patterns would be damaging for all concerned.

Because the A and B parents believed that they could not effectively decrease prognostic uncertainty, they described most protective strategies as serving little purpose other than disturbing the normal flow of family activities and relationships: "You can't change nothing in a family- because if you change for one, what's going to happen to the rest of us?" This belief meant that the parents in the A and (to a somewhat lesser degree) B families were willing to take certain protective "risks" in order to better normalize the leukemia experience for their families. Mr. and Mrs. A's view was that:

> You don't know what's going to happen tomorrow, so she (the leukemic child) has got to enjoy herself...She can't enjoy herself if we protect her

> She enjoys herself if she does things...Sometimes we would worry that she might get hurt, but she'd have a good time.

"Keeping things the same" meant that rough-housing between the leukemic child and older brothers was permitted, and that the ill child was encouraged to participate in active sports activities:

> We tried to let her do what she wanted to do, you know, to try things (like)...riding the bike. (Chuckles) There was a lot of falling down. You know, she scraped shins and stuff, but... (Shrugs shoulders and laughs).
>
> She kept up with her figure skating. And she played soccer. We asked Dr. X, and he said she could do it, so we just watched her and she was okay.

The siblings in the A and B families agreed that "keeping things the same" was a beneficial strategy for their parents to have adopted in response to the leukemia diagnosis. However, they described its most valuable contribution as the fact that it had encouraged the maintenance of "equality" among the siblings. Given the finding from the literature that jealousy is one of the most common responses of children to chronic sibling illness (see Appendix A), it is not surprising that the children interviewed for this study all identified "special" treatment of any one sibling as an undesirable and potentially disruptive parenting strategy:

> If you treat someone different than they start to feel different...like they're not part of the family anymore...and before you know it, they're starting to act different, too.

In the three subject families where sibling "equality" had been largely maintained over the remission period, the children identified this characteristic as a major positive attribute of their families: "One of the things I like best about my family is that nobody's treated special. We're all the same- well, not really the same, but like equal." In the fourth family where preferential treatment of the ill child was part of the parents' protective strategy, the children all identified discriminatory "spoiling" of one or another sibling as a "not-so-good" aspect of their family.

The A and B parents were also concerned about the impact of "special" treatment of the leukemic child upon sibling relationships and family interaction, expressing apprehensions similar to those outlined by Boone and Hartmen (1972), Fostel (1978), and Poznanski (1973). These parents worried that preferential treatment of their ill children would result in spoiled and unmanageable long-term survivors, resentful siblings, and divided families:

> If you treat someone as if they're dying and then

> they don't, you've got one awful spoiled kid on your hands ...And those other two (the siblings) sure notice that, too. And before you know it, you've got two other spoiled ones in the opposite way-rebellious, resentful.

In order to prevent these damaging long-term consequences, Mr. and Mrs. A and Mr. and Mrs. B worked hard to maintain "normal" expectations and discipline for their leukemic children, even when this was emotionally difficult for them:

> She got just as much hell as anybody else when she didn't do her job.
>
> If she did something wrong, she got a licking for it -or she was sent to bed, the same as everybody was treated...That was hard...You know, you had to really hold yourself back from saying, "Well, we'll let her get away with it this time."...That was hard, but I think we succeeded.

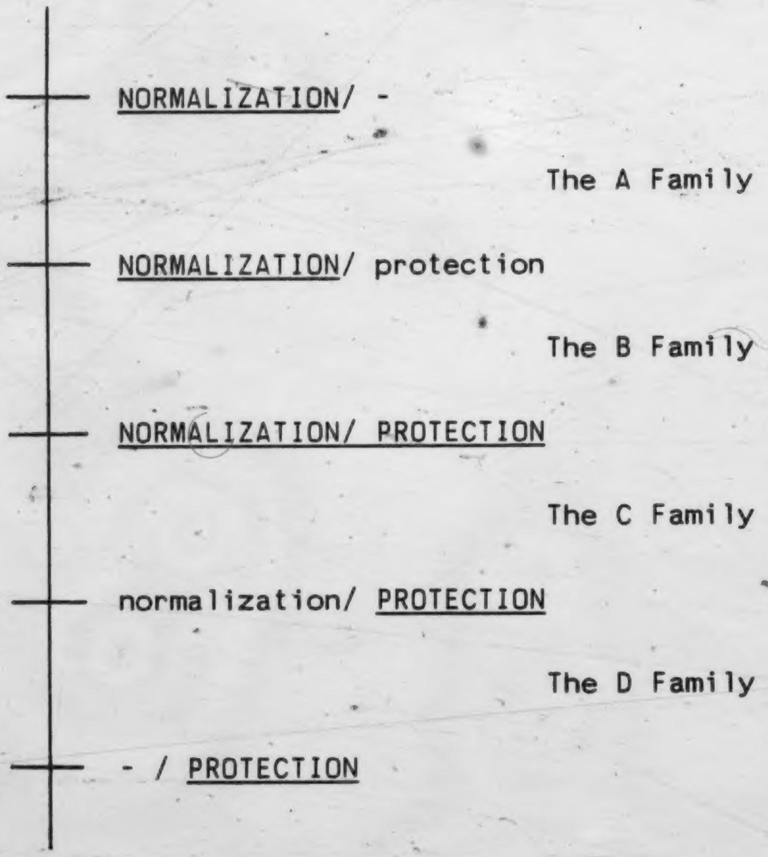
While the A and B parents concentrated primarily upon normalization-related coping strategies in living with long-term childhood leukemia, they acknowledged protection concerns and made use of some protective strategies. For example, while the A's encouraged their daughter to keep active in various sports, they initially sought the doctor's advice on these suggestions and then checked the child over for bruises after each activity session. While they did not expressly forbid her brothers from getting physically rough with her, "we (the brothers) just understood that you had to be kind of gentle with her". Although no specific dietary or vitamin regimes were used by the A family, the father stated he regularly exhorted his daughter to "Just eat, eat- that's good for you...Like steak...I said, that's good for you." For the B's, family camping holidays continued after the diagnosis of leukemia, except that now they stayed close to the city and slept in heated cabins rather than their tent-trailer:

> I think we did it sort of unconsciously, because we didn't really realize that we'd changed at all. Until later, when you start to realize that you have no up-to-date camping equipment and stuff that's sort of gone by the wayside eight years ago.

In reference to the normalization versus protection continuum depicted in Figure 8, it can be seen that the A and B families are not plotted at the extreme upper end of the scale. These two sets of parents continued to acknowledge and respond to some protection concerns, even while emphasizing normalization. It is probable, however, that some parents would respond to the same crisis by virtually ignoring protection concerns altogether in their attempts to "keep things normal" for the ill child and family. Examples of such responses could be refusing treatment (because it might make the child sick), going

Figure 8

The Families' Normalization Versus Protection Balance Points


NORMALIZATION/ -
The A Family
NORMALIZATION/ protection
The B Family
NORMALIZATION/ PROTECTION
The C Family
normalization/ PROTECTION
The D Family
- / PROTECTION

from doctor to doctor to find a "better" diagnosis, and ignoring suggested treatment-related measures which inconvenience the ill child or other family members. In order to carry out these strategies for any length of time, affected parents would have to have a powerful ability to deny the true nature of the disease and its implications. To distinguish this response from the denial-as-avoidance ("not talking about it") strategy to be discussed later in this chapter, the hypothesized primary coping response of extreme normalizers will be referred to as "maladaptive denial". This form of denial is termed maladaptive because its logical consequence would be a heightened risk to the health and life of the leukemic child, due to inadequate parental protection.

The "Protectors"

The experience of the C and D families in living with long-term childhood leukemia was dominated by the parents' efforts to actively protect the ill child from illness and death. The parents in both families had strong reservations about the abilities of the present health-care system to "cure" their children. Both sets of parents handled their anxiety about their children's uncertain prognoses by evolving elaborate systems of "protective" rules and rituals. Alterations in family life-style and interaction patterns were seen to be the necessary price for ensuring the survival of the ill child. As stated by Mrs. D:

> Although he (the leukemic child) had a hard time of it for the first while with that diet and those pills, at least he'll be around to make up his own mind when he's eighteen, whether he sticks with this or not. Not like the others (the four other children who had been diagnosed with leukemia within several months of her son)- they're all gone.

In devising their protective strategies, the parents in the C and D families made use of very different approaches and philosophies. The C parents emphasized the use of rules restricting the activities of their ill child. Mr. and Mrs. D responded to the leukemia diagnosis by introducing fundamental life-style changes into the lives of all family members. While both sets of parents expressed normalization concerns about the siblings and their responses to the ill child, they used two opposing approaches in acting upon these concerns.

The C parents' protective strategies included hypervigilance and the curtailment of activities for their leukemic child. Their hypervigilance was directed towards the detection of any indication of infectious disease (especially chicken pox) in the children with whom their ill son interacted:

> I used to take them (the children) to church and then look around for spots...even when they (the doctors)

> said I didn't have to worry about it anymore.
>
> I used to take them to public swimming...and I watched for spots and for fever (in the other children at the pool) ...I'd ask, "Is your child in a lot of mosquitoes- I notice a lot of spots on him."

The activity curtailments arranged by Mr. and Mrs. C included discouraging their leukemic son from active sports or playing out of doors, restricting activities such as swimming and running through sprinklers, and prohibiting tussling with older brothers. As Mrs. C remarked, "I really felt protective of him. I could have just cuddled him up and kept him close." The C family vacations were limited to activities which were perceived to be "safe" for the ill child and which took place within a reasonably short distance from the major treatment centre. According to one sibling:

> We wanted to stay in places where we could get close to hospitals...and we also didn't want to go to places where he couldn't do anything...(so) we kept away from fishing 'cause of the cold off the water and sitting in the boat for long.

Mr. and Mrs. C did express some normalization concerns about their family:

> We were really trying to keep their (the siblings') lives as normal as we possibly could. We didn't want this to overshadow their entire lives...We didn't

want them to resent Mike.

These parents attempted to meet their normalization concerns by keeping life "as normal as possible" for the well siblings through limiting major life-style changes to the ill child and the parents. This approach meant, however, that while the siblings were encouraged to "keep things the same" (i.e. keep up former activities), their ill brother was now treated in an openly different (i.e. unequal) manner from them. The well siblings noted the "special" food the ill child received to entice him to eat, the "special" presents to compensate for the activity restrictions, and the "special" parental attention. Their reactions to the perceived "special" status of their ill brother were strong and will be discussed in the next section, under Consequences.

Mr. and Mrs. D solved the problem of incorporating normalization concerns into a predominantly protective orientation in a very different way from that of Mr. and Mrs. C. The D's focussed upon "equality amongst the siblings" as the critical aspect of normalization, instead of "keeping things the same". This focus meant that when they began considering the major life-style changes required by the Laetrile and herbal treatment methods, they decided that all family members would be expected to alter their diet and activities in order to avoid making the leukemic child "feel different". This approach also allowed them to feel that they actively protecting themselves and their other children from cancer, which they suspected was primarily caused by carcinogens in their food and soil. Lamontagne (1984) and George et al. (1980) confirm that individuals with an internal locus of health control (such as the C and D parents) typically rely heavily upon vigilant or active strategies in their coping responses to illness.

The specific protection strategies instigated by the D family were varied and wide-ranging. First, in accordance with the recommendations of the Laetrile clinic, all refined, processed, artificial, and deep-fried foods were eliminated from their diet, and "natural" foods were substituted. In order to obtain adequate amounts of organically grown vegetables, the family began to grow its own- and eventually, established a market garden business. Goats were raised to ensure an ongoing source of raw goat's milk. Every child took a variety of vitamin and mineral pills, and was expected to regularly consume "healthy" beverages such as carrot juice. Due to concerns about "jarring" the ill child's brain, all the D children were withdrawn from hockey and other contact sports and instead enrolled in the local bowling league. Activities that involved the consumption of "junk food" (i.e. church picnics) were discouraged, and new friendships with "health-food type people" were formed. However, in order to demonstrate some flexibility in accommodating their children's tastes, Mr. and Mrs. D did permit "junk food" (i.e. cake, potato chips, and pop) to be eaten on special occasions such as birthday parties.

Although not demonstrated by any of the four sample families, the logical extension of the protection orientation, if taken to its extreme, would involve the total neglect of family or normalization concerns in order to mobilize all resources for the protection of the ill child. This response is defined by Boone and Hartmen (1972) as "benevolent over-protection". According to these researchers, parents who see their children as handicapped or seriously chronically ill may attempt to minimize and compensate for the difficulties associated with their health conditions by "over-protection, over-indulgence, and permissiveness" (p. 268). However, the long-term consequences of these actions can be as or more serious than the original illness itself, as they include decreased self-esteem and self-control in the ill child, resentment among the well siblings, and disturbed parent-child relationships. It is therefore hypothesized that an extreme emphasis upon protecting the ill child, demonstrated by the use of "benevolent over-protection" strategies, would pose a direct threat to the well-being- and perhaps the very existence of- the family unit of the chronically ill child.

Strategies Used by Both "Normalizers" and "Protectors"

Two parental coping strategies were described by members of all the families. These were "pulling together" and "not talking about it". "Pulling together" involved two main aspects: practical assistance and emotional support. The parents all made use of relatives, friends, and health-care professionals for practical aid such as baby-sitting and advice:

> My sister took the kids whenever we had to go into the hospital. She was just like a second mother to them.
>
> Oh, the kids knew, when they came home, they just went straight over to the neighbour's.

The parents also used a variety of sources for emotional support, but relied especially upon their spouses. As one wife put it:

> It really pulled us together- the two of us, I mean. When we were first married, we used to have fights about all sorts of silly things. But after she got sick, those things just didn't seem important anymore.

The fact that these families all had a variety of support sources available to provide practical and emotional assistance (as outlined in Chapter 4) may have played a large role in their overall positive adaptation to the leukemia experience over time.

Of all the coping strategies described by the various family members, "not talking about it" was by far the most frequently mentioned and, apparently, the most widely used. "Not talking about it" included three inter-related sub-strategies. These were: "limited sharing/ seeking of information", "not thinking about it", and "keeping feelings to oneself". In general, minimal communication about the disease and its implications appeared to be the norm in the subject families. The limited sharing of disease-related information with the siblings was a major parental strategy in three of the four families.

"Not talking about it" was usually described by the parents as a protective measure designed to limit sibling anxiety and potential family disruption. As one father stated:

> I think if the parents are nervous when the kid's sick, if you're jumpy, then you put it on the kids, too, you see. So don't mention those things- it's all over. I don't think we mentioned it after we talked to the kids once and said she got sick.

The children in this family were given no information about the ill child's treatment regime or prognosis. In another family, the well siblings were not told of the diagnosis until one month after it was given to their parents. Nonetheless, long before this time, the disruptions in routine and obvious anxiety of the parents had convinced the children that something terrible (and probably life-threatening) was happening to their brother. However, the possibility of death was not addressed by the parents with these siblings for another year, until one of the well children mentioned his earlier fears. The parents in this family were surprised that the siblings had guessed the true nature of the leukemia prognosis, and stated their children had "managed to pick up a lot more than you would think".

According to the parents, the major purpose of "not talking about it" was to "protect" the well siblings from worry and fear. Featherstone (1980), however, points out that another important reason parents may withhold disease-related information from their children is that difficult questions from inquisitive children could threaten their own ability to avoid acknowledging distressing disease realities. For example, one set of parents explained that they were initially reluctant to discuss leukemia with their well children because they feared facing questions which they had not been able to answer for themselves:

> They (the siblings) never mentioned it and we didn't want to mention it- that we were afraid Mike was going to die...(But) that's what people say, that you have to discuss with your kids even, you know, what you think of life after death and whatever happens

> and all those feelings. And I thought, "How can you pass on to them until you get it figured out in your own mind?" It'd be easy if you had all the answers, and they asked a question and you answered it. But sometimes you wish you could keep some of this stuff inside.

In other words, the parents found that the "limited sharing of information" strategy was a necessary concomitant to the often useful "not thinking about it" strategy.

The "not thinking about it" coping mechanism did not appear to be used by the parents as an attempt to negate the problem of the leukemia (the classic defense mechanism of denial). Instead, they employed it as a "deliberate attempt to avoid thinking about a threat, which (was) nonetheless regarded as real" (Cohen & Lazarus, 1979). These two types of denial can be differentiated by the titles "denial-as-negation" versus "denial-as-avoidance" (Lazarus, 1981). Parental statements showing the use of denial-as-avoidance included the following:

> You can never really forget about it (the leukemia) ...but I just put it way back, into the very back of my mind.

> Well, we were all very busy with the hockey and the soccer and everything else the kids were involved in, and we just didn't have time to sit around moping

about it.

Besides directly limiting the amount of leukemia-related information provided to their children, the interviewed parents enforced their "not talking about it" policy in a number of ways. Sibling questions were in effect discouraged by the use of brief answers which often made little sense to the children, and by the restricting of "talking about it" sessions to information-exchange only. There is little evidence (except in the B family) that these parents ever encouraged their children to share their feelings or perceptions related to the leukemia experience with them. When sibling feedback was solicited in the rare family conferences about the issue, it was almost exclusively in the form of answering, "Do you have any questions?":

> The whole family would get together once in awhile and talk about what was going on. But they (the parents) never went in deeply- like all the feelings and stuff. We just talked about, like how he was doing and if we had any questions.

Unfortunately, as several of the siblings pointed out, during times when they had the most need for information, they were sometimes too frightened or confused to know what questions to ask:

> They (the parents) started telling us about that (the leukemia), and I got a bit scared, and I didn't ask

> questions. I guess I sort of stayed away from the family. I didn't ask many questions at all.

As described in the following section, the siblings made use of a variety of "not talking about it" strategies which complemented those of their parents. For example, the parental "limited sharing of information" was matched by the siblings' "limited seeking of information". As they got older, the siblings asked even fewer questions of their parents, and increasingly relied upon the "not thinking about it" and "keeping feelings to oneself" coping responses. The sibling coping focus upon their own emotional responses to the leukemia rather than upon pragmatic aspects of the situation was the main distinguishing characteristic between the parental and sibling coping strategies.

Comparison of Parental and Sibling Coping Strategies

Lazarus and Folkman (1984) describe coping strategies as falling into two main categories: problem-focussed or emotion-focussed. These two authors define problem-focussed strategies as those oriented "toward doing something to solve the problem" (p. 44). Emotion-focussed strategies are directed towards "regulating the emotional response to the problem" (p.150). In the present study, the problem-focussed coping strategies were those which involved active attempts to protect the ill child and/ or to normalize the situation for the family. Because of leukemia's indefinite prognosis, however, emotion-focussed coping strategies were also widely used by the families to minimize the anxiety associated with long-term uncertainty. In comparing the strategies employed by the interviewed parents and their well children to cope with long-term childhood leukemia, it is important to realize that while the parents' made use of both problem and emotion-focussed responses, the siblings relied almost exclusively upon emotion-focussed approaches. The major parental coping strategies are summarized in Table 6; those of the siblings are outlined in Table 7.

Several general points need to be considered when examining Tables 6 and 7 and comparing their contents. First, the siblings participated in parental coping strategies such as "keeping things the same" and "life-style changes" which were adopted as family responses to the leukemia. These, however, are not included as "sibling coping strategies" as they were not initiated by the siblings themselves. Second, according to Lamontagne's (1984) framework which differentiates between protective (or active) and avoidant stress responses, "normalizing" coping strategies would be described as avoidant (and therefore emotion-focussed). However, these strategies are defined here as

Table 6

Major Parental Coping Strategies

Coping Strategy	Problem (P) or Emotion (E) Focused	Primary Users (Parents of A, B, C, or D Families)
Protecting:		
- Vigilance	P	C, D
- Restrictive Rules	P	C, D
- Life-style changes	P	D
Normalizing:		
- Keeping things the same	P	A, B, (C)
- Maintaining equality among the siblings	P	A, B, D
Pulling together:		
- For practical assistance	P	A, B, C, D
- For emotional support	E	
Not talking about it:		
- Limited sharing of information	E	
- Not thinking about it	E	A, (B), C, D
- Keeping feelings to oneself*	E	
Giving meaning*:		
- Re-defining the situation	E	
- Having faith in powerful others	E	A, B, C, D
Worrying	E	A, B, C, D

*Less frequently described strategies

( ) = Less use of strategy than with other cited parents

Table 7

Major Sibling Coping Strategies

Coping Strategies	Problem (P) or Emotion (E) Focused	Primary Users (Siblings from A, B, C, or D Families)
Not talking about it:		
- Limited seeking of information	E	All. Use ↑ with ↑ length of remission
- Not thinking about it	E	
Keeping feelings to oneself	E	
Seeking information/ reassurance from parents	E	All-used in Alarm & early Vigilance (& in middle childhood)
Worrying and ruminating	E	All-mostly used in Alarm & Vigilance
Giving meaning:		
- Redefining the situation	E	Some use by all
- Having faith in powerful others	E	
Normalizing:		
- Joking and teasing	E	A, D
Protecting:		
- Vigilance	P	C, D
- Protecting survivor from others	P	A, B, (C), D

( ) = Less frequent use of strategy

problem-focussed because they were directed towards solving one of the two major sets of concerns arising from the leukemia diagnosis. Third, it is clear that some parental coping strategies designated as problem-focussed also served important emotion-focussed purposes. For example, it is debatable whether the main effect of the D's laetrile and herbal treatment methods was in actually protecting the ill child from relapse, or in making the child (and family) feel protected. It is therefore acknowledged that the following classifications of coping strategies are somewhat arbitrary, and that many strategies served dual purposes.

Parental Coping Strategies

As can be seen from Table 6, the parental coping strategies included both problem-focussed and emotion-focussed responses, and contained components of each of the five conceptual categories listed in Table 1. The main problem-focussed strategies used by the parents were "protecting", "normalizing", and "pulling together for practical assistance". All the parents made use of the "pulling together" strategy, while the primary users of the various "protecting" responses were the C and D's. All parents made some use of "normalizing", but only the A and B parents emphasized both its "keeping things the same" and "maintaining equality among the siblings" sub-strategies. The major emotion-focussed parental coping mechanisms were "pulling together for emotional support", "not talking about it", "giving meaning", and "worrying and ruminating". These strategies were used by all the parents, although to differing degrees. Within the broad "not talking about it" strategy, the parents made most use of the "not sharing information" (with the children) and the "not thinking about it" (through denial or avoidance) substrategies.

The "giving meaning" coping strategy subsumed a variety of approaches occasionally mentioned by the parents. Parents gave meaning to their experience in a number of ways. During the reappraisal process, some parents chose to actively re-define the nature of their child's leukemia:

> After five years off treatment, the Cancer Society says she is cured- and I've been starting to say so, too...I feel that she is cured now, that there's not going to be a relapse.

Other parents took consolation from comparisons with children more seriously ill than their own:

> We talked to parents of kids... (who) weren't doing so well. And all of a sudden, you learn to think that, "Gee, we're better off then they are."...I guess you learn to be grateful for small mercies.

Several parents described how the leukemia experience had taught them to re-focus from long-term to more short-term family and individual goals, and to take pleasure in celebrating even minor milestones:

> We used to make a big deal out of it- if we would go from once a month (clinic visits) to (every) three months, and then from three months to six months, we'd go out and have a fancy supper. You know, a little celebration.

Sibling coping strategies

With the one exception noted in Table 7, the coping strategies used by the well siblings were exclusively emotion-focussed. Due to their dependent position in the family and limited understanding of the situation, the siblings had little opportunity to become actively involved in promoting normalization or protection outcomes. Because they could do little to alter the circumstances with which they were faced, the siblings concentrated instead upon regulating their own understanding of and response to these circumstances. Koch-Hattem (1986) also noted that affect dominated the descriptions of post-diagnostic life changes given by siblings of pediatric cancer patients in remission.

The most widely used sibling strategy was "not talking about it", which included the sub-strategies of the "limited seeking of information", "keeping feelings to oneself", and "not thinking about it". The siblings' "limited seeking of information" response was the flip-side of their parents' "limited sharing of information" strategy. The siblings' pattern of limited information-seeking evolved as they matured and the disease situation changed. In the early period after the diagnosis, the siblings asked questions about the leukemia that were primarily reassurance-seeking in nature: "Is he/ she going to be alright?" "Is he/ she going to die?" As the sense of immediate threat passed and the siblings got older, more general information questions came to the fore: "Why does he/ she have to get back pokes?" "Why can't he/ she play outside with the rest of us?" Once the early acute stages of the disease had passed, the number of questions asked by the siblings, though never large, decreased significantly. By early adolescence, most of the well siblings stated that they almost never discussed the leukemia or its implications with their parents- and that they could not remember the last time they had asked them a question about it.

The decline in sibling information-seeking may be attributable to several factors. First, after the initial crisis of the diagnosis, some of the siblings felt they were protecting their parents from further anxiety by not mentioning the disease. The unspoken family rule that leukemia was not to be talked about was quickly internalized. Second, as the children moved from alarm to vigilance to relaxed vigilance in their re-appraisals, their questions seemed less pressing to them: "If everything's going okay, what's there to ask?". Third, as they matured to greater independence, the siblings tried to deal with more of their personal concerns and difficulties by themselves: "I keep more to myself about everything now."

While the siblings did not express a great number of leukemia-related fears, typically they kept these questions and concerns to themselves:

> I just think about it by myself where I am, or go to a quieter place. Go for a walk or something.
>
> Well, I don't ask my mom any questions about it...I keep to myself. Just keep to myself and don't say anything unless I'm asked.
>
> I bike out to this place where I have this fort, and just stay there for awhile to think about it and stuff.

Parents were often unaware of their well children's leukemia-related concerns. Sharing feelings was not encouraged in the families, except to some degree in the relatively open B household.

As the complement to "keeping feelings to oneself", the "not thinking about it" sibling coping strategy was employed in two different ways: defining the ill child as "cured" or avoidance and distraction. A significant number of the siblings had convinced themselves that because the leukemic child was "cured", there was no longer any potential problem to worry or think about: "Now that she is fine and everything, I can just deal with it because nothing is going to happen." "Because the doctors have pronounced her cured... like I can sort of rest back in ease that she's okay." The children's definitions of their leukemic siblings as "cured" were often challenged by the new questions and perspectives which occurred to them during their early adolescent years. When doubts about cure and long-term prognoses surfaced, the siblings used the "not thinking about it" ploys of avoidance and distraction to prevent their fears from becoming overwhelming:

> I just try not to think about it. I just say that it won't come back.
>
> It wasn't until a few years ago that it really hit me that gee, you know, my sister could have died there. But then I said to myself there's no use thinking about it.

At times when "not thinking" about the leukemia was impossible (such as during the Alarm and early Vigilance phases of re-appraisal), the siblings had typically responded by worrying and ruminating. Occasionally, this worry was expressed somatically with nightmares or stomach upsets. Some siblings responded by placing their faith in powerful others such as laetrile or the health care team: "I thought, "They're professionals- they know what they're doing."." For others, anxiety was decreased through normalizing the situation with jokes and teasing: "She lost her hair and she had to wear a wig and stuff, and she got some ribbing from us, which was par for the course." Re-defining the situation helped some siblings rationalize family changes which they found disturbing:

> Like I knew he was sick at the time, so it (the special treatment) didn't really bug me. I thought that he should be getting some special attention.

The only problem-focussed coping strategy described by the siblings was that of "protecting" the ill child. This strategy included being vigilant for any evidence of relapse or sickness in the leukemic child, as well as protecting them from the taunts (physical and verbal) of their peers. The need for "vigilance" over the ill child was especially emphasized by the children of the C. and D. families:

> Well, like he's pretty cured...but you've got to keep a wary eye out.

> The main thing you can do is keep on watching them... make sure they're still okay, that everything's going like they (the doctors) planned.

However, siblings from all four families were concerned about protecting the leukemic child from harassment and discrimination:

> Kids at school started bugging him because he had a hat on and not too much hair...If I caught up with them, then I'd beat them!

The siblings' use of the coping strategies outlined in Table 7 waxed and waned with their ongoing re-appraisals of the situation- and with their maturing. In general, the siblings' use of strategies that involved the parents for reassurance and information decreased over time, as they increasingly kept feelings to themselves and devised their own explanations for phenomena. Further, about half the siblings had difficulty discussing the evolution of their coping strategies with the investigator, because the presence of the leukemia was such a minor and/ or accepted part of their existence that "we just lead our normal lives". The combination of the parental and sibling coping strategies described in the foregoing section appeared to be generally successful in helping the siblings cope effectively with the long-term uncertainty of childhood leukemia.

Coping Consequences for the Siblings and their Families

Given the number of years and family life-events which took place between the time of the leukemia diagnosis and the study interviews, it is impossible to designate valid cause-and-effect relationships between coping strategy use and specific long-term outcomes. However, there were some general sibling responses which appeared to be logical manifestations of certain sibling and parental coping strategies. Some of these responses occurred across all families, and others were specific to either the normalizing or protecting families. These long-term sibling responses are summarized in Table 8, and discussed below.

Sibling Beliefs re Impact of the Leukemia Experience

As explained previously, all members of the four interviewed families had made extensive use of the coping strategies within the category "not talking about it": "limited sharing/ seeking of information", "not thinking about it", and "keeping feelings to oneself". These strategies were generally seen by the subjects to serve protective purposes for the individuals employing them, and for the family as a whole. Numerous authors have argued that such avoidance responses can serve adaptive purposes for families forced to live with chronic illness

Table 8

Late Sibling Responses to Living with Long-term Childhood Leukemia

Sibling Response	Siblings Showing The Response (From A, B, C & D Families)
Belief re: impact of leukemia experience on self and family:	
a) None or minimal	a) A, B ("Normalizers")
b) Noticeable	b) C, D ("Protectors")
Leukemia - related anxiety:	
a) None or minimal	a) Most siblings
b) Some	b) Those siblings in early adolescence and/or who had many negative leukemia reminders
Belief in value of family coping strategies:	
a) Best possible strategies	a) A, B, D
b) Improvements suggested	b) C - where equality among siblings not maintained
Increased family closeness	A, B ("Normalizers")
Sense of need for ongoing vigilance re: survivor	C, D ("Protectors")
Minimal Knowledge About Leukemia	All

and long-term uncertainty (Allan et al., 1974; Beisser, 1979; Fostel, 1978; Lazarus, 1981; O'Malley et al., 1979). These writers point out that avoidance responses which appear to represent a denial of illness may in fact be intended as an affirmation of health, hope, and life. Such avoidance strategies provide family members with mechanisms that can help prevent them from becoming bogged down in a morass of despairing speculation about outcome possibilities. Over the long-term, these "not talking about it" coping strategies had a number of consequences for the siblings.

Overall, the heavy reliance upon "not talking about it" coping strategies appeared to have achieved its purpose of avoiding undue distress for family members. The siblings did not display overt evidence of serious maladjustment, or, in most cases, of note-worthy ongoing anxiety. Most felt that their families' lives had not been seriously disrupted by the leukemia experience, and in fact agreed that their families had probably been strengthened and consolidated by it: "Things worked out really good. They worked out for the best."

In the normalizing A and B families, where change had been actively resisted and little evidence had existed of the disease's possible implications, some of the siblings felt that the leukemia experience had had no meaningful impact upon themselves or their families:

> Like everything was just normal...It was like nobody was overly worried. My parents were normal, and they just went about everything the same way. And our meals didn't change and our lifestyle, it didn't change at all.
>
> I don't think it has made any difference at all...We have learned to cope with it, and it's just that. We haven't really done anything about it.

The A and B parents also agreed that:

> There was minimal impact (on the well siblings)... (It was) just routine, more or less. It sounds terrible to say that something like that could be routine! (Laughs) But that's really the way it was, I guess.

The A and B siblings who felt that the leukemia had had some long-lasting impact upon themselves and/ or their families all agreed that this impact was slight and largely positive because it had brought their families closer together.

In the C and D families, the parents' emphases upon "protecting" their ill children had resulted in a variety of changes in their families. Not surprisingly, the C and D siblings believed that the leukemia experience had had a noticeable impact upon themselves and their families. As one sibling stated:

> We had to change. We had to take care of ourselves

> more because my parents had to be gone. We had to change to make everything better.

In the D family, the siblings noted numerous life-style changes after the leukemia diagnosis such as:

> We're more health-food-wise. Eat a lot more of that. We're more cautious about what we eat, and about the unpronounceables on the food labels.

According to the D children, such life-style changes were beneficial long-term outcomes because they improved the health and disease-resistance of all family members. As one declared, the family's health-food diet "saves me from the junk food". The C siblings gave less concrete examples than the D children about the long-term impact of leukemia upon their family. However, the effects they alluded to, such as increased family fighting and tension and continued wariness about "special" treatment of any one child, were largely negative. These effects appeared to stem primarily from the parents' failure to "maintain equality amongst the siblings".

Leukemia-related Anxiety Among the Siblings

Another sibling consequence related to the "not talking about it" coping strategy was little or no ongoing sibling anxiety about the leukemia or its implications. As noted earlier, the siblings tended to quickly assume their ill brothers or sisters were well or "cured", unless they were faced with concrete evidence to the contrary.

All siblings were in the Relaxed Vigilance stage of cognitive re-appraisal, meaning that overall they had few lingering doubts about the "cure". There were two groups of siblings, however, who demonstrated noticeably more doubts and long-term anxiety than the others. These were most of the young adolescent siblings (ages eleven to fourteen) and the siblings who had been exposed to significant ongoing evidence of the seriousness of the leukemia diagnosis.

Given the improved cognitive abilities which accompany the onset of adolescence, it is perhaps not surprising that the three siblings who appeared the most uncertain and worried when discussing the leukemia with the interviewer were between the ages of eleven and fourteen. These siblings were struggling to come to terms with the troubling issue of uncertainty which Cohen (1986) describes as dominating the long-term leukemia experience. Their younger siblings seemed less aware of the complexities of this issue, while the older teenagers appeared to have resolved the question to their own satisfaction.

In general, the interviewed siblings who had been exposed to little evidence of the seriousness of childhood leukemia (in the form of parental anxiety, family changes, and the witnessed traumatic procedures) had virtually convinced themselves of the leukemic child's "cure".

Furthermore, it appeared that they had not worried (or even seriously thought about) the disease for months or years prior to being interviewed for this project. On the other hand, those siblings who had been faced with numerous reminders of the life-threatening nature of leukemia, and who had therefore remained in Vigilance for extended periods, tended to express ongoing disease-related worries which they described as surfacing every few weeks or months:

> I don't know why I start thinking about it. I don't know. When I'm alone, nothing to do, just sit down and think about it, and then that's usually when I do think about it... about death. Like that the leukemia could come back, or he could get something else that could make him sick, or other things that could happen to my family.

Those siblings, then, who had been the slowest to enter Relaxed Vigilance were also least certain about their leukemic sibling's long-term prognosis.

Sibling Belief in the Value of Family Coping Strategies

With the exception of one family, the siblings all agreed that the normalization versus protection orientation adopted by their parents had been the best approach possible for coping with childhood leukemia. Furthermore, they generally attributed their family's positive long-term adaptation to the particular coping style used by their parents. When asked for advice for other families facing the crisis of a recently diagnosed childhood leukemia, the siblings' answers typically reflected their family's coping strategy use. For example, siblings from the "normalizing" A and B families believed that minimizing change and the ill child's "difference" within the family were the most important precursors for "coping well":

> What would I say? Don't change whatever you're doing. ~~Keep it the same way~~ as what you're doing, and just let her (the ill child) go do what she wants. I wouldn't say changes- no way, that's impossible, no.
>
> I think to make you (the leukemic child) get better, part of it is people treating you like you were before, cause that does, I think that does help the person.

Meanwhile, siblings from the protective D family emphasized the need for major life-style changes, including the use of laetrile:

> Advice? I don't know. Go to Mexico. Get some Laetrile. Start the diet. Like cut out all the junk food. And it's better if you can move to a farm and grow your own stuff, because then you know what's in them, and what you're eating.

While the siblings from the A, B, and D families could not think of any ways that their parents could have coped better with the leukemia experience, the C siblings had specific suggestions for improvement. These children had found it difficult to resolve their parent's early post-diagnosis anxiety and "special" treatment of the ill child with their reticence to provide adequate explanations for these responses. More importantly, however, these children had trouble accepting the idea that measures necessary to protect their ill sibling needed to be conducted at the expense of sibling equality. The primary importance of "maintaining equality amongst the siblings" was a point stressed by all siblings. When this principle was violated in the C family, the siblings responded vigourously:

> It made me mad...(I) cried (and) had some fights about it...Like we'd go, "Why is he getting all these presents? You shouldn't treat him like that!" Like, we'd just shout at them, you know..."You guys (the parents) are mean. You guys don't like us anymore and all you do is care about Mike." And other stuff.

Crain et al. (1966) suggest that when parents attempt to compensate a child with gifts and extra time for the losses associated with his/ her chronic illness, the well siblings can quickly "become handicapped in the race for parental attention". These siblings can then be expected to react to their reduced status with resentment, jealousy, and anger.

While the D parents had, like Mr. and Mrs. C, adopted a predominantly protective orientation towards coping with longterm childhood leukemia, their strategy of having all their children share in the changes and restrictions created for the ill child prevented the well siblings from feeling neglected or slighted. In fact, in this family the siblings perceived the differences between the treatment accorded them and their leukemic brother to be in their favour. The only jealousy mentioned in the D family was the leukemic child's envying of the more lenient dietary and medication regimes followed by his well siblings:

> Well, when I was on the strict diet, they would sometimes get a treat or something after. And I wasn't allowed to have anything. So sometimes I thought they were a little better than I was.

Increased Sense of Family Closeness

All the siblings of the "normalizing" A and B families described an increased sense of family closeness as a positive long-term consequence of the leukemia experience. A and B family members described themselves as "close" before the diagnosis, but even closer after:

> I would say that overall, although it's a terrible thing that you wouldn't wish on anybody, that it

> really has served to pull our family together.

For the A and B's, the family focus of their normalization strategies served to pull them together. This consequence was aided by the general lack of sibling resentment or jealousy of the ill child, due to the parents' emphasis upon "maintaining equality amongst the siblings". In contrast, none of the C or D siblings suggested that their families had become closer as a result of the leukemia experience. In fact, in the C family, the impact of the leukemic child's "special" treatment appeared to have had the opposite effect on family unity.

Despite reports of an increased sense of family closeness, however, the subject families' extensive use of "not talking about it" coping strategies seemed to have isolated the well siblings from each other, their parents, and (to a lesser extent) the ill child. Evidence of individual isolation within the sibling subsystem was provided by the siblings' reports that they did not discuss leukemia-related thoughts or feelings amongst themselves, ~~and only occasionally with~~ the long-term survivor:

> I don't know how they (the other siblings) felt, because...we never did talk much to each other. So we didn't talk about feelings between one another. We might talk to our parents, but we never talked to one another.

Evidence of the isolation of the well siblings from their parents was provided by the fact that in only one family did the parents ever sit down with any of their children specifically to discuss the feelings aroused by the leukemia. The overall picture was one where family members worked together (often unconsciously) to preserve the delicate balance of minimal information and emotion sharing upon which their mutual avoidance strategies depended. The unintended consequence of this approach, however, was that to a large extent, each well sibling was left alone with fantasies and concerns based primarily upon inference and ignorance.

Ongoing Sibling Awareness of the Need for Vigilance

When asked whether there was anything that could be done to keep their brothers' or sisters' leukemia from returning, the well siblings from the two normalizing families stated either that little or nothing could be done. In the protection-oriented C and D families, however, the siblings stressed repeatedly the need for the doctor and themselves to maintain vigilance over the leukemic child, although it was not clear what they (the siblings) were "keeping a wary eye out" for. According to the D siblings, their brother's survival depended not only upon maintaining protective vigilance, but also upon the continuation of the laetrile and health-food related life-style changes to which they attributed his original "cure".

Minimal Sibling Knowledge about Leukemia

An important sibling consequence of the "Not talking about it" coping strategies was a marked lack of knowledge about leukemia and its consequences. This finding coincides with those of Burton (1975), Spinetta (1981), and Allan et al. (1974), who found that a significant number of well siblings (often over half their sample) lacked even the most fundamental knowledge about their brother or sister's chronic illness. In the present study, only one long-term survivor was able to give a reasonably detailed description of leukemia. The siblings' understanding of the disease was limited, vague, and sometimes erroneous. Not surprisingly, the siblings with the least complete understanding of the disease came from the family where the "Not talking about it" policy was most strictly observed (the A family). The definitions of leukemia provided by the siblings in this family (ranging in age from fourteen to twenty-four) included the following:

> Definition- well, it's a serious cancer. Definition of leukemia. I couldn't really give you a definition because I don't really know that much about leukemia. So I know it's a cancer, you know, but I just don't know that much about it...I really don't know. What is it?

> Definition? Oh, it's in your blood and ...you lose hair, I guess, at the top, and...I don't know. I just know it's in your blood. I think.

One adult sibling in the A family was surprised to learn about the possibility of relapse:

> Can it come back, then? I didn't know. Can it come, I mean I should probably know this stuff, but can it come back?

In the D family, the two siblings differentiated between leukemia and cancer, describing leukemia as a "less serious" disease than cancer. Based upon the teachings of their parents, these siblings were the only ones who had any ideas about possible "causes" of leukemia, suggesting "chemicals" in "junk" foods and radiation from the sun and nuclear testing. One of the B children described leukemia as something you could catch like a cold. Typical definitions of leukemia given by siblings from the B, C, and D families were:

> It's a cancer of the blood. And it's where there isn't enough white blood cells to get rid of diseases and so they have to try to build him back up.
>
> All that I know so far is that it's a cancer of the blood. (And what is cancer?) Well, I know it's bad. I don't know too much else about it.

Notably, none of the siblings demonstrated any awareness of the long-term implications of relapse.

The high level of overall sibling ignorance about the leukemia and its implications was an logical outcome of the "not talking about it" coping strategies employed in the name of protecting oneself and others from undue emotional distress. This outcome, while perhaps psychologically non-taxing, may leave the well siblings poorly prepared to face a variety of possible future leukemia-related difficulties such as relapse and second malignancy.

Summary

In Secondary Appraisal, each set of parents was faced with the decision of how best to balance their normalization against their protection concerns. Two of the sets of parents chose to emphasize normalization over protection, while two focussed primarily upon protection. The major normalizing strategies were "keeping things the same" and "maintaining equality among the siblings", while "protecting" included "vigilance", "restrictive rules", and "life-style changes". The parents used these and other problem-focussed coping strategies to deal with the practical issues associated with their children's leukemia. To help decrease the anxiety related to the uncertain prognosis, the parents also made use of a variety of emotion-focussed coping strategies, especially "not talking about it".

The coping strategies used by the siblings were almost exclusively emotion-focussed, and complemented those of their parents'. The most frequently used sibling coping strategies were "keeping feelings to oneself", the "limited seeking of information" and "not thinking about it". The overall long-term consequence for the well siblings of the combined parental and sibling coping strategies was that they demonstrated no obvious signs of psychosocial maladjustment that could be related to the disease experience. Over half expressed little or no present anxiety about the leukemia or its implications, while the remainder described some ongoing disease-related concerns. Most had very little knowledge about the disease, although almost all had some awareness of the uncertain nature of its prognosis. The siblings all agreed that their families' coping strategies were the best possible for dealing with childhood leukemia, except when these strategies had resulted in the perceived "special" treatment of the ill child.

Chapter 7

Conclusions

Summary and Conclusions

A number of important findings have come out of this study, and are recapitulated below.

Social Support

The families interviewed for this study defined themselves as coping either "well" or "fairly well" with living with long-term leukemia in a child. A major characteristic shared by these families was the extensive social networks of friends and relatives each used for practical and emotional support during difficult times in the leukemia experience. Numerous authors identify adequate social support as a crucial component of positive family adaptation to chronic childhood illness (Holaday, 1984; Kazak & Marvin, 1984; Krulik, 1980; Schilling et al., 1984; Spinetta & Maloney, 1978; Venters, 1981). The major sources of parental social support in this study were found to be similar to those reported by Morrow, Hoagland, & Morse (1982)- spouses, then friends and relatives, followed by the health-care team. Religion was a further "very" important to "fairly" important source of emotional support for three of the four families.

Relationship between Appraisal and Coping

This study focussed upon the process of sibling and family adaptation to living with long-term childhood leukemia. "Balancing the demands" was identified as the core category around which this process was organized. "Balancing the demands" was a decision made by the parents early after the diagnosis which was determined by their appraisals of the leukemia situation and which, in turn, determined much of their coping response to that situation. In other words, most differences in parental coping strategy use could be traced to differences in parental situational appraisals.

The causal relationship between appraisal and subsequent coping apparent in this study is consistent with the theories of Lazarus (1966), Lipowski (1970), Mechanic (1961), and others. This relationship is most extensively described by Lazarus and Folkman (1984), whose stress, appraisal, and coping framework was used to help organize and conceptualize the results of this project. Several gaps in this framework as it applies to children within the context of their families were identified. For example, besides the situational and personal factors which influence parental appraisal and coping, sibling appraisals and reappraisals were also affected by four unique factors. These factors (described below under Re-appraisal) were the parental control of information, age-related cognitive limitations, reliance upon inference, and the tendency to assume wellness.

Balancing the Demands

During Primary Appraisal ("What is going on here?"), the parents' identification of the threats posed by the leukemia to the well-being of their ill child and family resulted in the development of both protection and normalization concerns. The parents' "balancing the demands" decisions between normalization and protection represented their attempts to effectively reduce and tolerate the long-term uncertainty associated with childhood leukemia. A similar balancing response to chronic disease-related uncertainty has been noted in patients with rheumatoid arthritis (Weiner, 1975) and multiple sclerosis (Davis, 1973). Weiner found that the press of the "physiologic" and "normal activity" imperatives upon the arthritic force them into a precarious balancing act which is characterized by severely limited options and a great deal of situational uncertainty. For parents of leukemic children, the "balancing the demands" decision is also characterized by extremely high stakes and subsequent marked anxiety.

Locus of Control

The parents' "balancing the demands" decisions were determined primarily by their perceived locus of control. Locus of control refers to whether people view health outcomes as largely due to their own actions (internal locus) or to the manipulations of fate and powerful others (external locus). Many studies of stress responses have shown locus of control to be a major determinant of coping strategy use (Anderson, 1977; George et al., 1980; Lamontagne, 1984; Parkes, 1984; Rothbaum et al., 1979). In general, these authors suggest that individuals with an internal locus of control "will be more likely to use strategies focussed on altering the stressful situation, whereas externals will be more likely to adopt palliative coping strategies" (Parkes, 1984, p. 656). In the present study, the internally oriented parents felt compelled to emphasize the protection of their ill child and accept some compromise of their normalization concerns. The externally-focussed parents delegated most of their protection concerns to the doctor and instead emphasized normalization issues. The siblings' locus of control designations over the leukemia outcome typically echoed those of their parents'.

Re-Appraisal

The parental and sibling re-appraisals of the meaning and implications of the leukemia progressed through a number of discernible stages. There were important variations, however, between the parental and sibling re-appraisal time-lines, and between the two major variations of the sibling time-line. These differences could be attributed to the four major characteristics of sibling appraisal mentioned earlier. First, siblings were largely dependent upon their parents for information and interpretations relevant to the leukemia. Second, because of their cognitive limitations and their parents' reluctance to share disease-related information, the siblings relied largely upon inference and imagining in devising their private explanations for the situation. Third, the siblings tended to assume that the leukemic child was well or cured, unless they were presented with concrete evidence to the contrary. Siblings who were exposed to the fewest distressing reminders of the leukemia's presence and significance were the quickest to assume the disease's disappearance and the most convinced about the certainty of "cure". Fourth, the siblings typically re-evaluated their assumption of certain or almost certain leukemia survival in early adolescence, when the development of abstract reasoning abilities raised new questions in their minds.

Normalization

Some type of normalization strategy was adopted by every family in this study in order to prevent potentially damaging repercussions of the leukemia experience upon their psychosocial well-being. Normalization has been identified by various authors as an effective coping response to living with chronic childhood illness (Anderson & Chung, 1982; Futterman & Hoffman, 1973; Holaday, 1984; Krulik, 1980; Schulman, 1983; Wolfensberger, 1972). Typically these authors define normalization as the "continuation of usual patterns...and familiar routines" (Futterman & Hoffman, p. 136). In the present study, however, normalization was found to have two key components. Parents generally described it as "keeping things the same", whereas the siblings emphasized the aspect of "maintaining equality amongst the siblings". This latter aspect may have been overlooked in the earlier research because all previous studies in this area focus solely upon parental definitions and use of normalization strategies.

As can be seen in Appendix A, jealousy and resentment are the most frequently cited sibling responses to chronic childhood diseases and handicaps. In the present study, the only sibling resentment noted was in the family where the leukemic child had received preferential treatment, thereby destroying sibling "equality". Numerous authors have speculated that the root of sibling maladjustment to long-term childhood illness may be in parental pre-occupation with the sick child and neglect of the well ones (Cairns et al., 1979; Lavigne & Ryan, 1979; Peck, 1979; Teitz et al., 1977; Turk, 1964). The importance of "maintaining equality amongst the siblings" in promoting positive sibling adaptation to long-term leukemia was apparent in the present study.

Some authors refer to normalization strategies in terms such as "emotion-focussed", "avoidant", and "palliative" (Anderson, 1981; Lamontagne, 1984; Parkes, 1984). While its emotion-focussed functions are acknowledged, in this study normalization is referred to as primarily "problem-focussed". This designation is used because normalization concerns are deemed to represent major potential leukemia-related problems which the family must prevent or overcome in order to cope well with the situation.

"Not Talking about it" as Denial

The most frequently used emotion-focussed coping strategy among both parents and siblings was "not talking about it", which included the sub-strategies of the "limited sharing/ seeking of information", "not thinking about it", and "keeping feelings to oneself". The purpose of this strategy was the avoidance of thinking or communicating about the potentially devastating reality of childhood leukemia. This coping response could therefore be described as a form of denial.

Considerable controversy is evident in the family crisis literature about the function and impact of denial. Classical psychoanalytic theory defines denial as a pathological defense mechanism (Haan, 1980; Vaillant, 1971). Many recent authors, however, argue for its adaptive utility, especially in the face of long-term uncertainty (Allan et al., 1974; Fostel, 1978; Lazarus, 1981; O'Malley et al., 1979). Cohen and Lazarus (1979) differentiate the defense mechanism of denial (an attempt to negate the problem) from the functionally useful response of avoidance (the deliberate attempt to avoid thinking about a problem, which is nonetheless accepted as real). Denial-as-avoidance forms an integral part of many parental normalizing strategies (Anderson, 1981; Featherstone, 1980), and did serve some adaptive purposes for the interviewed families.

Various authors have noted that while denial-as-avoidance can be very effective in meeting short-term goals such as anxiety reduction, it may result in certain long-term difficulties (Desmond, 1980; Featherstone, 1980; Lazarus, 1981). In the present study, the "not talking about it" strategy appeared to have some undesirable consequences for the siblings such as disease-related isolation within the family subsystem and minimal knowledge about leukemia. Similar sibling consequences from the parental use of avoidance with chronic childhood disease have been noted by Anderson and Chung (1982), Burton (1975), Carandang, Folkins, Hind, & Steward (1979), Gogan et al. (1977), and Powazek et al. (1980). "Poor" or "restricted" family communication has been noted in many studies of the family response to childhood cancer (Binger et al., 1969; Cain et al., 1964; Futterman & Hoffman, 1973; Kaplan et al., 1976; Peck, 1979; Wold & Townes, 1969), and has often been hypothesized as a major contributor to sibling maladjustment to the situation. It is possible that "not talking about it" may be useful primarily for the parents of leukemic children, while turning out to be somewhat problematic for the well siblings who lack external avenues for information and support gathering.

Parental coping strategies

The parents used a variety of problem-focussed and emotion-focussed strategies in their efforts to cope with long-term leukemia in their children. These strategies are similar to the "effective" parental coping strategies described in the literature (and summarized in Chapter 2), except that the study parents relied upon "not talking about it" rather than open family communication. The parents emphasized either protection or normalization problem-focussed coping strategies, depending upon their earlier "balancing the demands" decision. All parents made use of similar emotion-focussed coping strategies and the "pulling together" problem-focussed response.

Sibling coping strategies

With the exception of "protecting" the ill child, sibling coping responses to the long-term leukemia were exclusively emotion-focussed. This finding is similar to the results reported by Koch-Hattem (1986), except that less use of "not talking about it" and more active reassurance, information-seeking, and emoting was evident in her sample. The differences between the two sets of findings may be due to the fact that the disease was a much more immediate reality for Koch-Hattem's sample, with only six to thirty-six months having elapsed between diagnosis and study interview. In the present study, sibling coping responses were found to be quite limited and to show less variety than those of their parents. However, their use was influenced by parental appraisal and coping responses.

Sibling Consequences

The major coping outcome for the siblings was that, with a few exceptions, they displayed little long-term anxiety and no obvious signs of maladjustment that could be related to their experience with long-term leukemia.

Similar findings were reported by Gogan et al. (1977), although these authors suspected that their retrospective method may have caused their subjects to appear more adjusted to the situation than they really were. In the present study, the two factors of being an early adolescent and numerous earlier reminders of the leukemia's seriousness were found to contribute to higher anxiety levels about the disease prognosis. Preferential parental treatment of the ill child was associated with more difficult sibling adjustment. Siblings from "normalizing" families reported an increased sense of family closeness since the diagnosis, while those from "protecting" families had an ongoing sense of the need to protect the leukemic child. Overall, the siblings had a minimal understanding of the disease or its implications.

Limitations of the Study

1. Retrospective studies are always weakened by the need to rely upon memory re-call. Important facts and perceptions of interest to this investigator may have been forgotten or re-interpreted by the subjects in the many years between diagnosis and project interview.

2. Several facets of the interview situations may have affected subject responses. First, in two families, the parents requested that the initial interview be a family interview (although not all family members were

present in either case). Beginning the interviews in this way may have influenced how the children described their experiences in later interviews. Second, privacy was difficult to achieve in two of the homes, due to their crowded conditions. This circumstance may have adversely affected subject openness in these two families. Third, one father was unavailable for interviewing due to prior work commitments.

3. The data for two of the sub-categories ("Having faith" and "Re-defining the situation") are less thick then that upon which the remainder of the categories and sub-categories are based. Less attention was inadvertently paid to these two sub-categories than to the remaining categories by the investigator during the interviews.

4. The presence of anxiety or psychosocial maladjustment was measured by the investigator only though inference, and not by any specific instrument. However, such instrument use is not appropriate in a qualitative project with such a small sample.

5. Due to social desirability, some of the interviewees may have described their families and their responses in a more positive (or more negative) light than was actually the case.

6. With the length of time passed between diagnosis and interview, the hypothesizing of relationships between coping strategy use and long-term sibling outcomes is tentative at best.

Recommendations for Further Study

Several suggestions for further study in the area of the sibling (and family) response to living with long-term leukemia in a child are proposed:

1. Because of the factor-searching nature and small sample size of this study, a replication study using a similar approach should be conducted to validate, and if possible, expand upon the findings in this project. Comparing such responses with those of families defining themselves as "not coping well" would also be useful. Furthermore, quantitative analyses of the frequency of "good" versus "poor" sibling and family coping with long-term childhood leukemia would provide a valuable supplement to the qualitative understanding of the process of such coping.

2. While the re-appraisal time-lines and coping strategies of the parents and well siblings of long-term leukemia survivors were compared in this study, this knowledge would be more complete if it was contrasted with similar information about the perceptions and responses of the long-term survivor.

3. Due to the limitations imposed by the retrospective study method, a prospective study focussing on sibling (and family) appraisal and coping strategy use with long-term childhood leukemia should be conducted. This project could examine at several points in time the major issues identified in the current study. For example, family member perceptions and responses could be analyzed and compared at the time of the leukemia diagnosis, at several intervals (perhaps every six months) during active treatment, and then yearly (or bi-yearly) for a period of time after active treatment was discontinued.

4. Much of the available coping research focuses upon the responses of adults acting alone or in the context of other adults. Significantly less information is available about the coping of children, and that which exists often examines the child's responses in isolation from those of the family. Much more needs to be understood about the relationships between parental and child coping, and how this changes over time as the child matures.

Propositions

The purpose of a factor-searching study such as the present one is to begin the theory development about a particular situation, which later relation-searching or association-searching research can refine, expand, and test (Diers, 1979). Reasonable theoretical propositions based upon the findings of the present study are the following:

1. Those families whose members will be most prone to long-term adjustment difficulties to living with childhood leukemia will be those who responded to the diagnosis by adopting extreme normalization or protection orientations (involving the virtual or total neglect of the other set of concerns).

2. Sibling anger and resentment towards the leukemic child will be most evident in families where the parents were unable to (or did not see the need to) "maintain equality amongst the siblings".

3. Siblings will tend not to resent family changes or protective restrictions instituted after the diagnosis of childhood leukemia, as long as those changes are not perceived to represent "special" treatment of the ill child.

4. On the "degrees of balance" continuum between normalization and protection, a family's position after the diagnosis of childhood leukemia will tend to gradually move towards the middle (representing a balance of the two sets of concerns) as the remission progresses and the threats posed by the leukemia seem less imminent.

5. In families who respond to childhood leukemia by adopting an extreme emphasis upon normalization (while ignoring most or all protection concerns), the main parental coping strategy will be denial (i.e. negation of the disease). This denial represents a threat to the survival and physical well-being of the leukemic child.

6. In families who respond to childhood leukemia by adopting an extreme emphasis upon protection (while ignoring most or all normalization concerns), the main parental coping strategy will be "benevolent over-protection" (as described by Boone and Hartmen, 1972). This reaction represents a threat to normal interaction patterns within the family, and thereby to family unity and cohesiveness.

7. After being informed of the leukemia diagnosis (and some of its implications), children (especially pre-adolescents) soon re-define their ill sibling as well or "cured", unless presented with concrete evidence to the contrary (i.e. parental anxiety, witnessing of traumatic procedures). This contrasts with their parents' tendency to assume that the leukemic child's survival is only "possible" (as opposed to probable) for years after the diagnosis- and sometimes indefinitely.

8. Children (especially pre-adolescents) typically base their appraisal of the seriousness of their siblings'

leukemia more upon inference from parental reactions and witnessed events than upon descriptive information provided by others (usually parents).

9. The multi-faceted "not talking about it" coping strategy described in this paper can be expected to be the single most widely used family response to long-term childhood leukemia, because it effectively serves the purposes of both normalization and protection. Use of the "limited sharing/ seeking of information", "not thinking about it", and "keeping feelings to oneself" strategies promotes the appearance of "normalcy" within affected families, and allows each member to feel they are "protecting" the others (and themselves) from undue disease-related anxiety.

10. One consequence of the avoidant "not talking about it" strategy is to decrease anxiety, thereby improving functioning and morale in the face of long-term uncertainty. However, it also isolates the siblings within the family with fears and fantasies based largely upon ignorance and inference. A further consequence is the establishment of a family coping style of noncommunication which may be very counter-productive in later crises (i.e. relapse, death) where informed mutual support is essential.

11. Because of the prevalence of the "not talking about it" strategy in families with leukemic children, one can expect that the well siblings will have little knowledge of the disease and its implications, and that they may have a variety of misconceptions about it. One can also expect individual well siblings to be relatively isolated (in terms of leukemia-related thoughts and concerns) within their families, discussing these issues to some degree with their parents, to a much lesser degree with the ill child, only rarely with outsiders, and never with each other.

12. Parents who express an internal locus of control about their child's acute leukemia can be expected to emphasize protection over normalization concerns in their coping efforts. Parents who express an external locus of control can be expected to emphasize normalization over protection concerns, as they feel there is little constructive they can do to "protect" their ill child, other then to follow the doctor's orders.

Implications for Nursing Practice

The findings of this study have a variety of implications for nursing practice. First and foremost, the results show that siblings and their families can cope effectively with the long-term uncertainty of "cured" childhood leukemia. In fact, some of the subjects interviewed for this study felt that, in the end, living with leukemia had proved to be a positive and consolidating experience for their families. Most of the subjects credited the specific coping strategies used by their families with their overall "positive" adaptation. The variety of normalization and protection strategies used by the parents demonstrates the range of effective coping styles possible in dealing with long-term childhood leukemia. It also points to the need for coping strategy advice to fit with the idiosyncratic situational appraisals of affected parents, if it is to be meaningful for them. Finally, the documentation of varying effective coping strategies points out the complexity of the relationships between coping responses and long-term outcomes, and acts as a note of caution against the providing of "pat" coping "prescriptions" for families living with childhood leukemia.

In considering the implications of this study for practice, it is important to realize that contrary to the opinion of certain of the subjects, positive adaptation to the leukemia may have occurred despite the use of certain family coping strategies, rather than because of them. As suggested by some of the interviewed parents, the most important variable determining coping outcome may have simply been the fact that relapse did not occur, thereby sparing the families from further tests to their coping abilities. The presence of further stressors could have strained the situation sufficiently to have more clearly demonstrated the weaknesses of the "not talking about it" coping strategies used by every family. While avoidance responses can serve adaptive purposes in helping family members continue to function in stressful circumstances, they also can isolate individuals within the family at a time when they most need each other. It appeared that the "not talking about it" coping strategies which helped the parents avoid dwelling upon unpleasant potentialities may have had the unintended side-effect of leaving the well siblings uninformed and unsupported in their coping efforts. In some cases, the siblings responded to their uninvolved status by assuming that, "No news is good news". In other cases, however, half-answered questions and unvoiced worries left the siblings feeling anxious, frightened, and confused for varying periods of time after the diagnosis.

It is widely accepted in the nursing literature that open, honest communication is a crucial component of effective family coping with crisis and serious disease (Share, 1972; Mott, Fazekas, and James, 1986). However, given their own uncertainties and need for stress reduction, talking openly with their children about leukemia may be a difficult task for parents (Featherstone, 1980). Consequently, nurses may have an important role in assisting parents to communicate more effectively with their well children about childhood leukemia. Suggested nursing interventions related to communication and other aspects of sibling and family coping with childhood leukemia are outlined below.

It is likely that parents will be better able to share information and feelings with their children if they feel adequately supported and informed themselves. The parents stressed the importance of having an "honest" doctor who could be trusted to answer their questions fully. They also appreciated the ongoing availability of a concerned health-care team who were willing to listen to their fears and discuss general issues related to childcare, school, family interaction, and so on. Some parents found the support of similarly affected parents (in the hospital or through organized parent support groups) to be valuable. One mother stated that the various scientific and research articles she requested from the health-care team had provided a source of background information to which she repeatedly turned for insight and guidance. Obviously, having a health-care team available with the time, knowledge, and listening skills to provide ongoing practical and emotional support for parents is one essential component of assisting families to cope effectively with childhood leukemia. A complementary second component is devising means to provide similar support sources for the well siblings, through the informed actions of their parents and others, including nurses.

In order to be able to promote positive adaptation in their well children, parents must be informed early of the need to talk openly with them about the meaning and implications of the leukemia diagnosis. Concrete suggestions and examples (preferably written, for ongoing reference) should be given, and sources of parental hesitation need to be identified and addressed. The interviewed parents had all been well informed of the importance of explaining the disease and its ramifications to their children. However, some had felt some uncertainty about how, when, and to what depth to carry these explanations. In order to decrease this type of confusion, nurses could use the results of this study to explain to parents how children typically perceive and assess their siblings' leukemia. This information could be supplemented with other research findings about children's typical understanding of sickness and death at various ages. As well as encouraging parents to study specially written reference books with their children, printed examples of age-appropriate explanations could be used to impress upon them the need to continue up-dating information as disease circumstances change and the siblings' cognitive abilities improve.

Parents were able to discuss some basic scientific information about the leukemia with their well children, but they had difficulty in talking about disease-related emotions or fears with them. In order to reduce the resultant emotional isolation experienced by the siblings, parents need to be encouraged to discuss feelings as well as facts with their children- and to realize that siblings will often not voice their inner concerns unless directly asked. Parents need to be aware that well siblings may express their worries in various unspoken ways, such as nightmares, stomach upsets, withdrawal, and behaviour changes. While initiating talk with children about their feelings on such an emotion-laden topic is not easy, parents may be encouraged by the following advice of two informants:

> (From a parent:) Knowing what we know now, about how they (the well siblings) were feeling at the time, it would have been better if we (the parents) had initiated talk about it (the possibility of death).
>
> (From a sibling:) They should have told us more. And maybe we could have helped out, instead of keeping every thing to themselves....After all, he's our brother, too.

Most of the siblings agreed that "open" communication was a desirable family characteristic, and a beneficial family response to the crisis of childhood leukemia. To conduct such "open" communication, however, parents may need specific information from nurses about effective listening and teaching techniques. If use of these techniques proved to be unduly taxing or threatening for some parents, similar information, support, and counselling functions could be carried out by a specially designated and trained nurse, or within the context of an organized sibling support group. The involvement of knowledgeable nurses (or others) could be especially useful to children at stressful times when parents are usually pre-occupied, such as while waiting in clinic playrooms listening to their siblings undergo painful invasive procedures. Special child-oriented group tours of clinic facilities, including opportunities to "practice" and ask questions about common diagnostic and treatment procedures, could help to make siblings feel informed, involved, and less frightened of what their leukemic brothers or sisters are experiencing (Cunningham, Betsa, & Gross, 1981; Kramer & Moore, 1983; McEvoy, Duchen, & Schaefer, 1985).

Finally, information from this study could be used to help siblings and their parents anticipate and understand the responses of various family members to living with childhood leukemia. It could serve as a means of emphasizing to parents the great importance children attach to "maintaining equality amongst the siblings", and to suggest various ways that this could be accomplished. The balancing-the-demands framework permits analysis of family coping strategies as predominantly protection or normalization-oriented, and provides rationales for concerns about extreme reactions noted in either direction. Most importantly, however, results from this study could be used to assure family members that by combining their caring efforts with the skills of a knowledgeable and concerned health-care team, it is possible for the ill child and his/ her family not only to "survive" childhood leukemia, but to grow and become strengthened through the experience.

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Appendix A

Types of Sibling Response to Chronic Childhood Illness, As Reported in the Literature

(Number of reviewed references citing response listed after each entry.)

1. Psychosomatic Disorders:
    a. Increased incidence of physical illness (8)
    b. Abdominal discomfort (2)
    c. Symptoms similar to the ill child's (2)
    d. Enuresis (4)
    e. Sleep disorders and nightmares (6)
    f. Eating disorders (3)

2. School Problems:
    a. General (4)
    b. School phobia (1)
    c. Decreased academic performance (8)
    d. Impaired cognitive abilities (2)

3. Emotional Problems:
    a. Increased anxiety (9)
    b. Jealousy and resentment (15)
    c. Increased fear of illness/Over-concern with own body (7)
    d. Guilt (8)
    e. Depression (5)
    f. Psychiatric problems (3)
    g. Separation anxiety (2)
    h. Negative self-esteem/poor self-concept (7)

4. Behavioural Problems:
    a. Social withdrawal and isolation (9)
    b. Fighting and aggressiveness (4)
    c. Worsened relationship with parents (2)
    d. Delinquency (3)
    e. Talk of suicide (1)
    f. General (4)

5. Positive Responses:
    a. Well-adjusted (7)
    b. Greater maturity and increased sense of responsibility (3)
    c. Increased tolerance and altruism (3)
    d. Assists with ill child (4)
    e. Improved relationship with parents (3)

Appendix B

Factors Influencing Sibling Response to Chronic Childhood Illness, As Described in the Literature

(Number of reviewed references citing factor
listed after each entry.)

1. Factors related to the ill child:
    a. birth order (4)
    b. age (3)
    c. gender (2)
2. Factors related to the sibling:
    a. birth order (10)
    b. age (7)
    c. gender (8)
    d. personality characteristics (4)
    e. typical response to stress (4))
3. Factors related to the family:
    a. family structure and roles (3)
    b. response of the parents to the illness (7)
    c. socioeconomic status of the family (4)
    d. ethnic/religious background of the family (4)
    e. typical family response to stress (4)
    f. family size(2)
    g. presence of other serious illnesses/stressors in the family (1)
    h. social isolation of the family (5)
    i. increased expectations of the well-siblings (5)

4. Factors related to the disease:
    a. type of illness(5)
    b. perceived social stigma of the illness (5)
    c. management requirements of the illness (3)
    d. visibility of the condition (5)
    e. ability to control the illness (1)
    f. prognosis (3)
    g. impact on body-image/self-concept of patient and/or sibling (6)
    h. severity of the illness (4)
    i. treatment side-effects (2)
    j. duration of the illness (1)
    k. stage of the illness (2)

Appendix C

Information Sent to Families.

Graduate Office,
Faculty of Nursing,
University of Alberta,
Edmonton, Alta. T6G 2G3
Mar.20, 1986.

Dear Mr. and Mrs.

First. let me say thank-you for agreeing to consider participating in my study. I realize my topic is a sensitive and sometimes difficult one, and I appreciate your willingness to allow your family to discuss it with me. My belief is that this study has the potential to contribute to better understanding and nursing care of families where a child has survived cancer.

For your information, I have enclosed the following documents:

1. A brief description of my study, including its objectives and method;
2. Sample consent forms for participating adult and child family members. (Please read these forms carefully, and have your children do the same. Child consent forms must be signed by you or your husband for your children who are under nineteen years of age. Extra copies of these forms are available so that we can each retain one for our records.)
3. Demographic data sheets- which ask for basic descriptive information about your family. (I will use this information to understand differences or similarities in the responses of the families being interviewed.) Please fill out this form. However, if there are any questions on the form you would rather not answer, please just leave them blank.

Please do not hesitate do call me at any time at my home number of 43 . (I am home most days and evenings.) Once again, thank-you for your attention to this matter. I look forward to speaking with you shortly.

Yours truly,

Kathleen M. Brett, R.N., B.S.N.

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NORTHERN ALBERTA CANCER PROGRAM

DESCRIPTION OF THE STUDY

The focus of my study is how children adjust to living with a brother or sister who is a long-term survivor of childhood cancer. It is well known that the period of diagnosis and treatment of childhood cancer can be a difficult time for all family members, including the siblings (brothers and sisters of the ill child). However, little is known about how children readjust over time as their ill brother or sister recovers and survives the disease. Studies of children with chronically ill or handicapped brothers or sisters show that some of them demonstrate long-term adjustment difficulties. It is not clear, however, whether these results have any implications for brothers and sisters of childhood cancer survivors, as there are important differences between the family experience of mental retardation or juvenile diabetes (for example) and that of childhood cancer. Therefore the primary aim of my project is to study how family members describe the response of the "well" brothers and sisters to living with childhood cancer for an extended period. I am interested in information about how it feels to be part of a family where one member has survived childhood cancer, how these feelings and reactions have changed over time, and what factors in the individual child, the family, and the situation appear to have affected the sibling's response. I believe this information will eventually assist nurses to provide more effective long-term care to families of children with cancer.

In order to gain insight into my chosen topic, I would like to interview all family members age ten and over (including the formerly ill child, if old enough) in selected families with a child who is a long-term cancer survivor. I would interview each person individually, on approximately three separate occasions. I would ask questions about the general family experience with childhood cancer (as it may have affected the brothers and sisters), and about the specific responses and perceptions of the brothers and sisters. In the first interview, I would concentrate upon clarifying my understanding of the family and individual experiences with childhood cancer. In the two subsequent interviews, I would ask more specific questions about the types of sibling and family responses noted and about the factors which may have affected these responses. For example, I would ask each person how family life has changed for them since the diagnosis of the cancer. I would ask whether there has been any change in the overall health, school performance, social activities, or behaviour of the brothers and sisters since that time. I would also be interested in knowing about things that have made life easier (and tougher) for the family in general and the siblings in particular since the time of diagnosis. Finally, I would ask about a variety of specific factors which earlier research suggests may affect a brother's or sister's response to the situation.

Should parents (and their children) agree to participate in the study, I will telephone them to describe the study again and to make arrangements to meet with them at their home (or some other convenient location). Before the visit, I will ask the parents to fill out the enclosed demographic data sheet. I will also ask them to carefully read the enclosed consent forms and discuss their contents with their children who are age ten and over. I will then meet with the parents at the pre-determined time to discuss the study and the consent forms, and to arrange individual interview dates. I will assure all family members that their particpation is strictly voluntary and that all information will remain confidential. I will also assure them that this information is being collected solely for scientific purposes and will not affect their relations with the Cross Cancer Institute in any way.

I anticipate the individual interviews to average about thirty to forty-five minutes (for children) and sixty to seventy-five minutes (for adults). Each interview will be tape-recorded in order to aid analysis of the conversations.

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UNIVERSITY OF ALBERTA FACULTY OF NURSING
INFORMED CONSENT FORM (CHILD)

Project Title: Childhood cancer as chronic disease:
The impact on siblings.
Investigator: Kathleen M. Brett, R.N., B.S.N.,
Graduate Office, Faculty of Nursing,
Clinical Sciences Building, University of Alberta,
Edmonton, Alberta. T6G 2G3 (Home phone no.:439-3874)
Supervisor: Dr. E. Davies, RN, PhD. (On sabbatical)
Alternate Supervisor: Dr. J. Morse, RN, PhD, PhD. Office phone: 432-6250

Purpose of the Study: The purpose of this study is to investigate the experience of children who are brothers and sisters (siblings) of long-term survivors of childhood cancer. It is anticipated that the results of this study will provide groundwork necessary for further research on strategies nurses and parents can use to help brothers and sisters adapt to this sometimes difficult situation. (For more detailed information, please see attached "Description of the Study" sheet.)

Consent: This is to certify that I, ______________________________ have given consent for my child/children:

______________________, ______________________

to participate in the research study outlined above. It is my understanding that I have given Ms. Brett permission to talk privately with my child/ children in our home (or another place agreeable to us all). I understand that three interviews, scheduled one to two weeks apart, will be held separately with each of the above-named children. I give permission for these interviews to be tape-recorded and for the tapes (with all references to family name removed) to be stored for three years in a locked cabinet (to permit possible further analysis at a later date by Ms. Brett). It is also my understanding that:

1. although the length of interview with my child/children will depend on his/her tolerance, the average interview will likely last about 30 to 60 minutes;
2. my child/children will:
   a. not have to talk about any subject he/she do not wish to discuss; and
   b. be assured that the content of his/her/their discussions with Ms. Brett will be held in confidence . However, my child/children will be free to discuss such content with myself, my spouse, or whomever he/she/they please(s);
3. I, my spouse, or my child/children will be free to terminate the interview at any time and to withdraw from the project at any time;
4. my child/children's name and the name of other family members will not appear in any research report;
5. my child/children may not necessarily directly benefit from participating in the study; and
6. my child/children may suffer some emotional discomfort if the interviewing revives painful memories and/or feelings. However, I realize that any interview will be stopped immediately if the child becomes upset. Constructive support by the researcher and/or the Nurse Counsellor of the Pediatric Oncology Team of the Cross Cancer Institute and/or Support Services of the Cross Cancer Institute will be available to any family member who requests and/or requires it.

I have been given an opportunity to ask whatever questions I desire of the researcher, and have had all these questions answered to my satisfaction.

Date:__________________ Signature:______________________________

Relationship to subject:______________________________

Investigator:______________________________

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NORTHERN ALBERTA CANCER PROGRAM

UNIVERSITY OF ALBERTA FACULTY OF NURSING
INFORMED CONSENT FORM (ADULT)

Project Title: Childhood cancer as chronic disease:
The impact on siblings.
Investigator: Kathleen M. Brett, R.N., B.S.N.,
Graduate Office, Faculty of Nursing,
Clinical Sciences Building, University of Alberta,
Edmonton, Alberta. T6G 2G3
(Home phone no.: 439-3874)

Supervisor: Dr. E. Davies, RN, PhD. (On sabbatical)
Alternate Supervisor, Jan.-June, 1986: Dr. J. Morse, RN, PhD, PhD. Office phone: 432-6250

Purpose of the Study: The purpose of this study is to investigate the experience of children who are brothers and sisters (siblings) of long-term survivors of childhood cancer. It is anticipated that the results of this study will provide groundwork necessary for further research on strategies nurses and parents can use to help brothers and sisters adapt effectively to this sometimes difficult situation. (For more detailed information, please see attached "Description of the Study" sheet.)

Consent: This is to certify that I, ________________________________ consent to participate in the research study outlined above. It is my understanding that I have given Ms. Brett permission to talk privately with me in my home (or another place convenient for me) on two or three separate occasions, scheduled one to two weeks apart. I give permission for these interviews to be tape-recorded and for the tapes (with all references to family name removed) to be stored for three years in a locked cabinet (to permit possible further analysis at a later date by Ms. Brett). I also give Ms. Brett permission to quote, report upon, or (if specifically requested) to tape-record social family interactions that she may witness or be involved in outside of the interview situation, provided that she requests specific verbal consent from me and/or my spouse prior to or during each such interaction. I understand that I will be asked to fill in a "Demographic Data Sheet", supplying basic information about my family. I give Ms. Brett permission to include relevant details from this questionnaire in her final report on the understanding that specific points which could identify my family will be removed. It is also my understanding that:

1. while it is estimated that the average interview will last about 60 to 90 minutes, I am free to terminate any interview at any time;
2. I will:
   a. not have to talk about any subject I do not wish to discuss; and
   b. be assured that the content of my discussions with Ms. Brett will be held in confidence by Ms. Brett. However, I will be free to discuss such content with whomever I desire;
3. I will be free to withdraw from the project at any time ;
4. my name and the name of other family members will not appear in any research report;
5. I may not necessarily directly benefit from participating in the study, and
6. I may experience some emotional discomfort from painful memories and/or emotions revived by the interviewing. However, I am aware that constructive support by the researcher and/or the Nurse Counsellor of the Pediatric Oncology Team of the Cross Cancer Institute and/or Support Services of the Cross Cancer Institute is available to me or any member of my family who may become upset during the interviewing involved in this study.

I have been given an opportunity to ask whatever questions I desire of the researcher, and have had all these questions answered to my satisfaction. I am aware that the researcher will send me a summary of the study results, if I so request.

Date:________________ Signature:________________________________
Investigator:________________________________

UNIVERSITY OF ALBERTA FACULTY OF NURSING
DEMOGRAPHIC DATA SHEET

Dear parent(s): Thank-you for filling out the following data sheet. The purpose of this sheet is to gather information about factors which may influence sibling response to childhood cancer. The information you provide will be regarded as confidential. (If you do not wish to answer a particular question, simply leave it blank.) Reference to this information in any published report will be made in such a manner as to prevent family identification.

1. Please list the first name, age, and grade of each child in your family:

Name	Age	Grade
______________________________	_____	_____
______________________________	_____	_____
______________________________	_____	_____
______________________________	_____	_____
______________________________	_____	_____

2. Please indicate your marital status:

_____Single
_____Married
_____Common-law
_____Divorced
_____Widowed

3. Please indicate your occupations:

Father's:____
Mother's:____

4. Do any extended family members live in the home with you?

_____Yes _____No

If so, how long have they lived with you? __________

5. What size of community do you live in?

_____Less than 5000
_____5000 to 10,000
_____10,000 to 20,000
_____20,000 to 50,000
_____50,000 to 100,000
_____More than 100,000

6. What is your family's ethnic background?

____

7. What is your family's religion?

_____Catholic
_____Protestant
_____Jewish
_____Atheist or agnostic
_____Other:_______________________________

8. Would you describe the role of religion in your family as:

_____Not important
_____Somewhat important
_____Important
_____Very important

9. Your total taxable family income last year was:

_____Less than $5000
_____$5000 to $10,000
_____$10,000 to $20,000
_____$20,000 to $50,000
_____More than $50,000

10. About your child who was diagnosed with cancer:

What year was your child diagnosed with cancer?
____
What type of cancer was it?_______________
Is your child currently off-treatment for his/her cancer?_____
If your child is off-treatment, how long has he/she been off-treatment?_______________

THANK YOU FOR YOUR INFORMATION!

-Kathleen M. Brett, RN, BSN

Appendix D

Pre-determined Guiding Questions
(Related to the factors and responses outlined
in Appendices A and B)

Note: In the questions below, X refers to the child with cancer and S refers to the sibling.

A. Types of Sibling Responses:

1. Psychosomatic disorders.
    a. Questions to S.:
    In the early days when X was sick, do you remember how your body felt? Can you describe those physical feelings to me? How long did they continue? (Can you remember any changes in how well you slept? Ate? How long did these changes continue?)
    b. Questions to the parent:
    How was S.'s health, generally, before X got sick? Did this change at all after X got sick? Can you remember any particular health problems S. developed? How long did they last? (Do you remember any changes occurring in S.'s sleeping habits? Eating patterns? Do you remember any bed-wetting starting?)

2. School problems.
    a. Questions to S.:
    Tell me about how things went for you at school after X first got sick. Did it make a difference for you there? (Did your grades change? Your attendence? Your attitude to school?)How are things now for you at school? Have things changed from when X first got sick?
    b. Questions to the parent:
    Did X's illness seem to affect S.'s school-work or school-life in any way? (After X first got sick, do you remember any changes in S.'s grades? Attendence at school? Attitude towards school? How long did you notice these changes continuing? What is the situation now?)

3. Emotional problems.
    a. Questions to S.:
    Can you describe your feelings in the early days when X first got sick? (Can you remember what thoughts you had about X at the time? About your parents? How did you feel about yourself at that time?) And as time went on, and X was home sometimes and getting his treatment sometimes, what were your feelings then? Do you have some of those same feelings still? Do you have them a lot? (Do you ever worry about getting sick yourself? Do you think that there is a difference in the way your parents treat you and X, because he once was sick?)
    b. Questions to the parent:
    Can you describe S.'s emotional response in the early days of X's illness? (How did he respond to you? To X? How did he/she seem to be feeling about himself/herself? Examples?) Did you have any concerns about S.? Have you noted any long-lasting emotional effects or changes in him/her? How would you describe his/her general emotional state today?

4. Behavioural problems.
    a. Questions to S.:
    (How did your friends respond when X got sick? Did your relationships with them change? How? Has it stayed that way? What about your relationship with the rest of your

family? When you get (or got) those feelings you were talking about before, what do you do (or did you do) to try and make yourself feel better?)

b. Questions to the parent:
Did you notice any behaviour changes in S. after X. got sick? Describe these changes. (Did you notice any difference in how S. got along with his friends? With the rest of the family?) How long did these behaviour changes continue?

5. Positive responses.

a. Questions to S.:
Can you think of any ways that X.'s illness has helped you become a better person? Has it been a good thing for you and your family in any way?

b. Questions to the parent:
Can you describe the ways, if any, that S. has benefitted from living with childhood cancer in the family? What positive changes have you seen in S. that might be related to this experience?

B. Factors that may affect sibling response:

1. Factors related to the ill child.

a. Questions to S.:
What difference (if any) did the fact that X. is older (younger) than you make to the way you felt about his/her sickness?

b. Questions to the parent:
(As above.)

2. Factors related to the sibling.

a. Questions to S.:
Did you notice a difference between the way you responded to X's illness and how your older(younger) siblings did? Was there a difference between how you reacted and how J.(sibling of opposite sex) reacted? Is there still a difference now?

b. Questions to the parent:
Have you noticed a difference in how the siblings older than X. responded to his/her illness as compared to those younger than X? Have you noticed a difference in the response of the girls compared to the boys? How has S.'s personality made a difference to his/her response (if at all?)

3. Factors related to the family.

a. Questions to S.:
Can you think of any things about your family that made you all react to X.'s illness the way you did? (For example, did your religion make a difference? Your neighbours and friends?) Describe to me how your parents reacted to X.'s illness- at first and now. How does their response make you feel?

b. Questions to the parent:
How do you feel your response (and that of your spouse) to X's illness has influenced S.'s response to it? What other things about your family may have made a difference? (Your religion? Family background? Friends and neighbours? Other problems to deal with?)

4. Factors related to the disease.

a. Questions to S.:
Was there anything about X.'s illness itself that made a difference as to how you felt (and feel) about it?

b. Questions to the parent:
Was there anything particular about X'x illness that seemed to affect how S. reacted to it? (For example- the hair loss and weight gain? Special fears about the word cancer? Awareness of the prognosis?)

Appendix E

Samples from Code

"Equality in the Family"

1. Mr. A: "Yeah, like I said, if you're sick that's fine, but everybody's got to be equal. You have to live with it."
Mrs. A: "Yes, for the rest of the family as well as her."
Mr. A: "Yeah, like we have seven of us here. And if one of us take different, what's going to happen to the rest of us?"

2. Sibling from A family: "No favouritism at all...My parents wouldn't do that. I know that for a fact. ...That's just the way they are...Everyone treated equal... If someone whoever did anything wrong, they got heck for it. And even if Carol, you know, did something wrong, they were sure to make sure that she got scolded or whatever. She had to do her homework and everything. ...My parents they would never, ever show favouritism, you know. It's always everyone's equal. Which is great, now that I think about it."

3. Sibling from B family: "What I thought was that she was just being treated the same...like, as with my other brother and me, she wouldn't be treated more favourably- like,quote- be more the favourite to mom and dad then we were...She's a member of the family. If there's something

that needs to be done, she pulls her own weight. And if she didn't..."

Interviewer: "She'd get it from you guys?"

Sibling: "Yeah- or mom and dad."

4. Sibling from B family: "That's what my mom and dad said when they found out that my sister had leukemia- that the doctor said to tell them not to favour her or anything, because she'll become the favourite and she'll never..grow out of it. So, she hasn't been favoured or anything. Nothing like that...Cause it's share and share alike, and everything for all... I like that there's no favouritism, and that it's always sharing."

5. Sibling from C family: "For awhile there I thought that maybe they were treating him special and that everyone was buying him things and that we were being left out. But after a while my parents explained to me that he does need that stuff 'cause he can't do the things that we do. And that he did have to go through the needles and the medicine and he needed something to cheer him up... For awhile I wouldn't accept that, but after awhile, I got to understand that things were becoming back to normal. He could do more things and they weren't buying him as many things and people weren't coming to visit to see him so much anymore. And everything just seemed to go back in place.... In that time, our parents tried to treat each one equally after they knew that he was pretty safe and

that."

6. Sibling from D family: "Like they don't treat him like he's a little baby. Or like he's a special one or anything. We get treated mostly the same. ...We're all, I don't know, equal... No one in our family's treated different than the rest."

7. Mrs. D: "Well, after he got sick, we pulled him out of the hockey. And then that didn't seem quite fair, so we pulled them all out."

8. Sibling from D family: "No one gets treated special...We're all pretty much the same. Since he had to start on the diet, we all did, too...Yeah, we're all equal."